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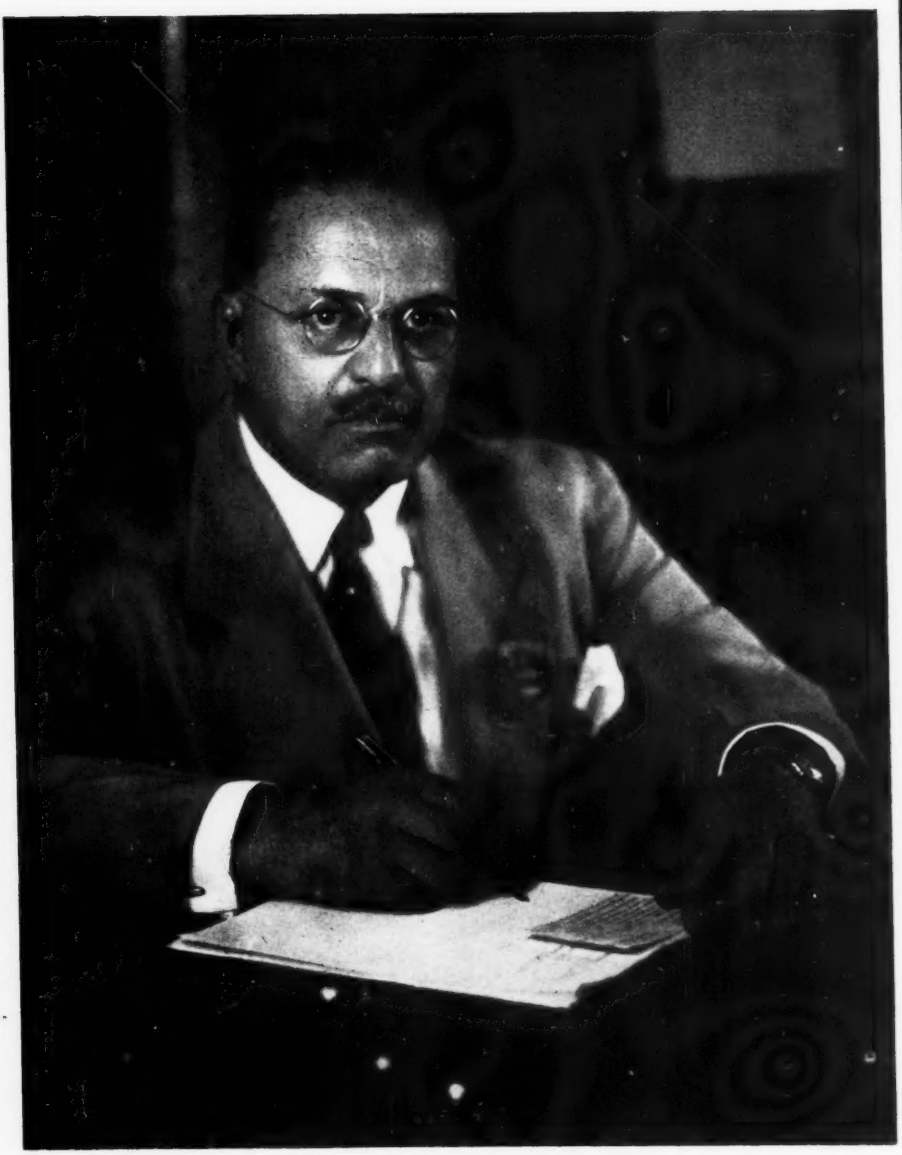

ERNEST SACHS

ERNEST SACHS

It is with the greatest possible pleasure that the Journal of Neurosurgery together with his many friends salute Ernest Sachs on the occasion of his seventieth birthday. That he has been one of the outstanding pioneers in our specialty very nearly contemporaneous with the trio of Cushing, Frazier and Elsberg is well known to us all. What is perhaps not so widely appreciated, however, is that unlike the men just mentioned Ernest Sachs set about his training to be a neurosurgeon in an orderly and planned manner, and thus doubtless became the first man to be regularly trained for this specialty with the idea of making it his life's work.

Being the son of a professor and the nephew of an outstanding neurologist it was natural for Dr. Sachs to look forward to a university career and, in medicine to have a particular interest in neurology. He was likewise influenced greatly in his earlier years by Professor George Parker, the biologist at Harvard. Later, in medical school, he was further stimulated by the teaching of Osler at Johns Hopkins and during his surgical internship in New York, by Dr. Arpad Gerster whom he, among others, has always regarded as an inspiring teacher and master surgeon.

Sachs' formal training in neurosurgery was with Sir Victor Horsley in London, and he thus learned of the trials as well as of the possibilities of this newest branch of surgery from the man who created it as a specialty. It was during his year with Horsley that he was asked by Cushing to come to Baltimore as his first resident, but under the circumstances he could not accept this offer which he greatly regretted. However, upon his return to this country he made a point of visiting Cushing's clinic on frequent occasions, and in the matter of surgical technic he quickly adopted more of the latter's methods than those he had seen employed by Horsley. It may be said, therefore, that he was a pupil of both.

After an early start in practice in New York Dr. Sachs was asked to organize a neurosurgical service at Washington University, St. Louis in 1911, and this service together with its teaching of students and training of young neurosurgeons has constituted his life's work. During World War I he was likewise acting professor of general surgery. This he was particularly fitted to do since he had had a broad experience in many surgical fields and had always kept up his interest in general surgery by having a weekly class in surgical diagnosis—a class which was frequently voted by the students to be the best course in the medical school. At the end of World War I he was made Professor of Clinical Neurosurgery, the first professorship with this title in the world.

In addition to membership in numerous medical, surgical and neurosurgical societies in this country Dr. Sachs has been made an honorary member of the Section on Neurology of the Royal Society of Medicine (England) and of the Deutsche Akadamie der Naturforscher. In 1943 he was president of the American Neurological Association, being the fourth

neurologic surgeon to hold this office. He was a founder member of the Society of Neurological Surgeons and its first secretary, later serving as president.

It has been one of Dr. Sachs' great contributions that he has trained a considerable number of young men and that all of those who are practising neurosurgery hold important positions. No less than seven of these hold professorships in this specialty.

His written contributions include two books: "The Diagnosis and Treatment of Brain Tumors" and "The Care of the Neurosurgical Patient," in addition to a long list of articles published in diverse medical, surgical and pathological journals. Some of his early experimental work was concerned with important investigations in connection with the functions of the optic thalamus in Sir Victor Horsley's laboratory. Later important contributions had to do with anatomic and physiologic studies on the eighth cranial nerve and studies on cerebrospinal fluid circulation.

His clinical papers have dealt with a great variety of neurosurgical subjects among which the diagnosis of brain tumors looms large, but there are also important observations on large series of cases of spina bifida and of internal hydrocephalus. He was quick to take up the use of the electrosurgical unit as a valuable adjunct in operations on the brain, and also detailed his experiences with direct radiation of brain tumors during operation, being the first person to employ this method. Another important contribution was concerned with the results and technic of subpial cortical resections for Jacksonian epilepsy.

But this tribute to Dr. Sachs is not meant to be concerned solely with his various honors and written contributions, valuable as many of the latter may be. Rather, we salute some of his personal characteristics which have always stood out so prominently to those who knew him best. From the professional standpoint there are perhaps two things which make their impress more than any others, namely, his diligent, continual concern with the teaching and training of young men, and secondly his unwearying and devoted attention to every detail connected with his patients—preoperative, at the operating table, and during their entire postoperative period. Students always had ready access to him as his office door was always open. Like many great teachers, he was ready to learn from them as well as to teach them.

So far as patients are concerned nothing is too much trouble for him if they can be benefited. He visits sick patients several times a day and never hesitates to come to the hospital at night to learn at first hand about any possible complication. All patients are at all times given the benefit of possible operative relief, often when their situation looks hopeless and when the result might affect his operative mortality adversely.

He has a healthy skepticism regarding newer methods and technics, yet does not hesitate to adopt them when convinced of their value. His discussions at medical meetings are always frank and to the point in spite of the fact that they might entail personal criticism.

Such an appreciation as this would be quite incomplete without a word concerning Mrs. Sachs. In addition to the quite obvious, and perhaps unusual devotion between them, there is mutual inspiration and understanding to a rare degree, which in turn cannot help but be reflected upon all those with whom they come into contact.

Ernest Sachs has lived through nearly the whole era of modern neurosurgery. He helped to develop his specialty in its infancy and toiled with other pioneers through the days in which the rewards of long and exhausting hours were few and far between. He has accepted and utilized the newer methods and made the most of them, and his clinic has always been a mecca for men from all countries who have wished to see the best in neurosurgery. He has been an inspiration to a host of students as well as to his contemporaries, and has shown extreme fortitude and devotion to his work in the face of varied vicissitudes, including long periods of physical pain.

We pay this simple but sincere tribute to one who has done much to further the best interests of our specialty and to whom neurosurgery and neurosurgeons owe much.

GILBERT HORRAX

NEUROSURGERY

YESTERDAY, TODAY AND TOMORROW*

WILDER PENFIELD, M.D.

Department of Neurology and Neurosurgery, McGill University, and the Montreal Neurological Institute

(Received for publication October 29, 1948)

A MAN who would make an after-dinner speech may be forgiven if he selects a title that promises great and unexpected things. It is like a dinner ménu, printed in a strange language. The appetite of the unsuspecting guest is whetted in anticipation when he discovers that he is about to be served something described, for example, as *pièce de résistance* even if, in retrospect, he should come to realize that it was only hash.

Yesterday, Today and Tomorrow is therefore an excellent title for my address. It could mean that you were to hear the polished prose of Sir William Osler. He was a Montrealer, physician and pathologist at the Montreal General Hospital and Professor at McGill before he started on his academic Odyssey. During his last years at Oxford I was fortunate enough to move in his orbit.

Listen to him as he gave to the students of medicine at Toronto the master-word in Medicine:

> It is the open sesame to every portal, the great equalizer in the world, the true philosopher's stone, which transmutes all the base metal of humanity into gold. The stupid man among you it will make bright, the bright man brilliant, and the brilliant student steady.
>
> . . . And the master-word is *Work*, a little one, as I have said, but fraught with momentous sequences if you can but write it on the tablets of your hearts, and bind it upon your foreheads.[5]

As you see, work sounds inviting when you call it the master-word, the open sesame to every portal!

My title for tonight's address could mean that you were to hear tragic poetry like that of John McCrae:

> We are the Dead. . . Short days ago
> We lived, felt dawn, saw sunset glow,
> Loved and were loved, but now we lie,
> In Flanders fields.[4]

McCrae was a Montrealer, a physician and pathologist at the Royal Victoria Hospital. He was a common man like you and me, until he stood in a trench in Flanders and heard above him the rushing sound of the wings of death.

Permit me one more quotation from a man who was dear to us here in

* Dinner address, American Academy of Neurosurgery, Windsor Hotel, Montreal, September 22, 1948.

Montreal, Dr. Edward Archibald, member of the Society of Neurological Surgeons and the first to practise that specialty in Canada. Twenty years ago he brought William Cone and me to Montreal and then abandoned the field to us to do what we could with it, while he pursued his own brilliant career in thoracic surgery. He could really have done justice to the title I have chosen. But now, alas, I can only recall his words at the time of the opening of the Montreal Neurological Institute:

> The nervous system is one of the most difficult parts of a man's frame to understand. Few subjects in Medicine have demanded of investigators as great an intellectual capacity or as arduous a labour. And in that very fact lies the reason and the justification for neurological specialism. The earnest man *knows* that neurology demands his whole life. But his guerdon is great.
>
> To gather knowledge, and to find out new knowledge, is the noblest occupation of the physician. To apply that knowledge with understanding, and with sympathy born of understanding, to the relief of human suffering is his loveliest occupation; and to do both with an unassuming faithfulness sets the seal on the whole.[1]

So, I have given you the ménu of this address. You might say I have proposed the toast, and three Montrealers more eloquent than I have responded to it. We Montreal neurologists and neurosurgeons welcome you, our guests from far and near.

Now I must draw forth from my own literary oven the *pièce de résistance.* I do so with a sinking heart, for I know quite well that when you, honest men and women, have sampled it you can only pronounce it hash. Nevertheless, here it is—Neurological Surgery, Yesterday, Today and Tomorrow.

Yesterday, between 1870 and 1900, neurosurgery was born in England. Principles such as localization of function within the nervous system, asepsis, anaesthesia, were combined so as to make possible a new therapy. This was the period of advance in the basic science of our specialty.

Today, from 1900 to now, we see the evolution of the technique of the surgery of the nervous system, chiefly in the United States. It is a familiar pattern: basic science in Europe, applied science here; basic atomic research in Europe, the "atom bomb" here. I hope to indicate that the time has come for us to turn our attention to basic research as well as to its application.

Cobb Pilcher took as the title of his excellent presidential address to the Harvey Cushing Society less than a month ago: "Neurosurgery Comes of Age."[6] So it has, physically, if not intellectually!

Young surgeons who have learned to use the scalpel so expertly that they can take anything out of anywhere without a fatality, to cut the pathways of the currents of the intellect and leave a man who is still capable of walking, may be tempted to look upon the performance of the pioneers in the earlier period with unjustified contempt.

Elaboration of surgical technique is an important mechanical achievement. But beware of vainglory; for it may be that our intellectual maturity is yet far off, and to be acquired only after years of further pioneering.

While Edward Jenner, a country practitioner, was elaborating the hy-

pothesis that he hoped would control the world's worst plague, namely vaccination for smallpox, John Hunter wrote to him, "Don't think, try; be patient, be accurate." Twenty years later Jenner tried, with the result that a million lives were saved.

Today we seem to be in so great a hurry! Too often we listen only to the first part of Hunter's injunction: "Don't think, try."

Tomorrow? What of Tomorrow? I believe that a new day will dawn tomorrow and that in its light will be found an understanding of the nervous system. Mental as well as physical activity will then be recognized as a function of the brain, and neurosurgery and psychiatry will gradually lose the separate authority conferred upon them by ignorance until there remains only neurology. Neurology will then stand forth as a single discipline to which internist, psychologist, surgeon, chemist and physiologist will contribute.

But before the dawn of that new day applied science must wait for the development of basic science. The neurosurgeon stands now in a strategic position. He alone can solve certain problems in human physiology, but he must stop to heed John Hunter's words, "be patient, be accurate." He must perceive the basic problems which block our advance, must think physiologically, chemically, anatomically or pathologically. The type of early training he has received will probably determine the manner of his thinking, if he thinks at all. Each can approach some problem according to his own ability, his own training, and according to the cry that reaches his ears from suffering humanity.

Perhaps, after all, we need a longer perspective before considering neurological surgery. More than 2000 years before Christ the Egyptians had innumerable specialists. As Herodotus, the historian, expressed it, some undertook "to cure diseases of the eye, others of the head, others again of the teeth, others of the intestines, and some those which are not local."

For two millenniums these specialists continued to practise and to collect fees without making the slightest advance. Indeed, they steadily degenerated. The trouble was that the Egyptians made a fatal mistake. They wrote text books, the hermetic books. They made another and more serious mistake, and that was to believe that the text books were correct. So they forbade physicians, at peril of their lives, to depart in any way from the treatment prescribed in the hermetic books. It was a remarkable experiment. Science (and art, religion and education as well) were all frozen by rules which, when originally written, may well have been the best that could be drawn up. The experiment demonstrated that standardization can halt advance but that it does not in any way hinder retrogression.

The only worthwhile heritage that came to us from the Egyptians was through the writings of Hippocrates, who obviously borrowed from their learning. There was, however, another thing we inherited directly from them and that was the charm, the incantation, which every physician invokes today to banish evil from the drugs he uses. I refer to the R with crossed tail

which mysteriously heads all of our prescriptions. This is the utchat, the eye of Horus, who was the Egyptian god of health.

Few physicians, and reputedly no surgeons, know what they are doing when they write a prescription today! Perhaps the knowledge that they have called upon such an ancient and honourable deity to free their formulas of any curse should be a consolation to them. In any case, I like to think that somewhere, in a forgotten Egyptian heaven, Horus is chuckling about it. On the other hand, it may well be that he looks down upon this new generation of specialists with alarm, lest they too should come to worship the perfection of their own achievement and so standardize treatment and training that basic science would again be frozen.

You who sit here tonight know exactly when and how the Academy of Neurosurgery was founded. But what about your mistress—how much do you know about her derivation? I refer, of course, to Neurosurgery. I propose to enquire for you into her past.

The date of her birth is uncertain, and also the place of her origin. Neurology obviously gave birth to her sometime after the conception of localization of function in the central nervous system. But she is not to be recognized as an individual until there was developed for her new techniques and a new set of principles. Some of you may be thinking that mistresses are usually distinguished for their lack of principles. Not so neurosurgery.

The great Joseph Lister once operated upon a patient for a brain tumour which had been localized for him by David Ferrier. When the dura was opened the pressure was so alarming that the operator plunged his thumb into the protruding cortex. Clear fluid gushed forth, as though he had used the rod of Aaron. Autopsy, which was not long delayed, showed that he had missed the tumour by a fraction of an inch. That, however, was not the beginning of neurosurgery. Nor would it have been if the luckless patient had happened to be relieved of his trouble, for many of the basic principles on which the specialty had to be built were still unrecognized.

In 1884 Rickman Godlee, a nephew of Lister, removed a brain tumour in the operating room of the Hospital for Paralyzed and Epileptic in Queen Square. The localization had been made for him by Hughes Bennett, according to the theory of cerebral localization lately enunciated by Broca, Fritsch, Hitzig, Ferrier and Jackson.

But, again, this performance was not the beginning of neurosurgery either. The localization was accurate, the surgery faulty. The brave young Scot who laid himself upon the operating table that day died weeks later of an operative infection. But the event may be looked upon as the prologue to neurosurgery, the curtain-raiser.

Wilfred Trotter,[7] referring to this operation 50 years later, said,

> We shall not get a proportioned view of our event unless we pause a moment to call up the world in which it took place. In 1884 we find ourselves in the richest depths of the Victorian era, and the warm tranquil air of its impenetrable security closes round us. . . . The land belonged to its monarchs, the seas belonged to Eng-

land, and the firmament was unconquered though perhaps still a little vaguely claimed by the Church. In science the air was as yet unadulterated with inert gases, and matter still sat contentedly in its little hard globular and indestructible atoms.

There was a neurosurgeon who could write as well as operate!

Those who were to play the leading role in the birth of our specialty followed every detail of this operation. Sir David Ferrier and Hughlings Jackson stood anxiously in mask and white coat to see the tumour come out, and two young surgeons, Victor Horsley and William Macewen, were in the audience when the case was reported.

I suspect that the first formulation of essential principles of neurological surgery took place in the minds of these two young men during the extended discussion of Godlee's case. I can imagine them listening to the enthusiastic talk of Ferrier, for only the year before, in his Marshall Hall oration, he had concluded his discussion of experimental brain operations in mammals with the argument that the time had come for the beginning of neurological surgery. I am sure the young men would have sought out Hughlings Jackson, then 50 years of age. Jackson had delivered a series of lectures eleven years before on "The Diagnosis of Tumours of the Brain"[3] and two years before on "Localised Convulsions from Tumour of the Brain."[2] No doubt, also, there was much conversation with the indefatigable clinician, William Gowers, then 39 years of age.

At all events, Horsley, who had just begun a brilliant series of physiological experiments of his own, found time to report a series of ten human craniotomies in 1886, and two years later he reported with William Gowers the first successful removal of an accurately localised tumour from the spinal canal. In the same year Macewen reported twenty-one craniotomies for brain abscess with eighteen astonishing recoveries. Perhaps the secret of this success lies in his first case which was not included in the twenty-one. I fancy that it was on his return to Glasgow from the discussion of Godlee's operation in London that he diagnosed an abscess in the frontal lobe of a boy. The parents refused permission for operation, but he secured the autopsy, tapped the abscess and inserted the drain, thus rehearsing step by step the procedure which was to save many lives. From basic science he had advanced to applied science.

Neurosurgery had had its birth, but not yet its baptism. It was not named a specialty and the technique those surgeons used was so crude as to bring a blush of shame today to the cheek of any member of this Academy.

I have begun with discussion of neurology and neurological surgery in London and have referred to the evolution of certain basic principles which were indispensable to the beginning of our specialty. For the next stage in its development we must turn to the United States, where a group of surgeons elaborated the technique, thereby reducing mortality to a reasonable level.

Harvey Cushing, surgical resident at Johns Hopkins, went abroad in 1900. He found Horsley too busy with practice and politics to be interested in his training. In Berne he worked on a neurophysiological problem sug-

gested to him by Kocher, drifted back to Liverpool to watch Sherrington experimenting with anthropoids, and on returning to Baltimore asked Professor Halsted if he might specialize in the surgery of the nervous system. Halsted suggested that he might better take up something useful, like orthopaedics!

However, in 1913, when he left Baltimore for Boston, he had become a specialist, a neurosurgeon, and in 1920 the Society of Neurological Surgeons was founded. In the next quarter of a century the specialty came of age under the leadership of Cushing, Dandy, Frazier, Sachs, Mixter and others. This was the period of technical evolution.

Not all leaders in neurosurgery were primarily surgeons. Otfrid Foerster, of Breslau, was a neurologist who assisted Kutner, the Professor of Surgery, for a time until abruptly, in an upsurge of impatience, he seized the scalpel himself. As might be expected, his contributions lay in the field of neurological mechanisms, not surgical technique. Neurosurgery for him was an opportunity to study pathological physiology and anatomy. Clovis Vincent, likewise, was a neurologist who slipped directly into neurosurgery after assisting Dr. de Martel.

It is splendid for men to be trained in surgery and apprenticed to neurosurgeons until they can carry out every standard procedure. That may be the "strait gate" and the "narrow way" of the orthodox. But there are other ways of entering the "kingdom of Heaven." I am sure that the great god Horus would join me in a plea that certificating boards should not close the gate to those who choose another approach. Not everyone can have a perfectly balanced training. Leave a crack in the gate wide enough to admit those of us who have had only surgical experience without approved neurosurgical apprenticeship, and for those with apprenticeship but with little general surgery.

The fields in which advance will be made "are white already to harvest." Those capable of taking in that harvest are few indeed. If there must be standardizing tests, and I suppose there must be, let them be intellectual ones and leave loopholes in the rules of time of service. Remember that the Egyptians achieved remarkable things in medicine and then retrogressed through two millenniums of standardized specialization.

My advice to men entering the field is to learn to speculate reasonably regarding its mechanisms. When it is that you learn this, what time you spend at it, and how you achieve it matters very little. It is never too late. Close up the flower of your busy practice for a few months. It may be then that your surgical tree will bear good fruit, whatever its past history.

Some men think in anatomical terms, others neurologically, psychologically, pathologically, physiologically, even chemically. There should be no room in our cult for those who think only financially, socially or not at all. Let certification boards aim to keep these men out, if indeed there are any such seeking admission.

It is enough for us to learn to think about our problems in terms of some basic science and learn to operate with adequate safety. This brings great

spiritual reward (to which, fortunately, financial reward is usually added!). To use the quaint phraseology of Archibald, the "guerdon is great," and again: "To gather knowledge and to find out new knowledge is the noblest occupation of the physician. To apply that knowledge . . . with sympathy born of understanding, to the relief of human suffering, is his loveliest occupation."

It is a source of satisfaction, no doubt, to devise technical procedures and to invent new gadgets. But this, it seems to me, is nothing compared with the reward that comes to a man when he at length evolves a general principle from many isolated observations.

Such a deduction may come to him suddenly, like a flash, while he is talking. It may come late at night when he is alone, writing out his analysis of observations. It may come when he has turned his mind quite away from the problem. But when it comes it is the result of his own intellectual effort.

Osler was right, I suppose. The master-word, the "open sesame," is work, for work must precede and work must follow a generalization, but the work which follows is different.

Whenever scientific creation comes to a man, and however unimportant it may seem to others, it is for him an exciting experience that he will not forget in his lifetime. The task of testing and retesting his principle is not a task at all for him, for each time he finds verification of his thesis there will return to him something of the exaltation of that first excitement.

Finally, what constitutes the specialty of neurosurgery? A body of accepted basic principles, yes, and a group of technical and operative procedures which conform to these principles. But something more—the alluring promise of new discovery. I have already pointed out that this is what makes her a formidable mistress. It is the call which explorers feel I suppose; difficult to explain but powerful. Kipling called it an "everlasting whisper." "Something hidden. Go and find it. Go and look behind the ranges—something lost behind the ranges. Lost and waiting for you. Go!"

The promise of discovery may be found in other special fields as well as in the nervous system, but nowhere is the future so full of promise, the possibility of advance so unlimited, and the results of research so important to mankind.

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TANTALUM CRANIOPLASTY

REVIEW OF 106 CASES IN CIVILIAN PRACTICE*

EDWARD C. WEIFORD, M.D., AND W. JAMES GARDNER, M.D.
Cleveland Clinic and the Frank E. Bunts Educational Institute, Cleveland, Ohio

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THERE are sufficient experimental[5,8,17,37,38,40,50,51] and clinical data[2,10,12,14,16,42,47,53] on the repair of cranial defects with tantalum to justify the acceptance of this surgical procedure as the best cranioplastic method now available. There is adequate evidence of the relative inertness of tantalum imbedded in tissues,[5,8,9,40,41,44] its easy malleability, and its adaptability to varied use.[6,23,24,27,38,39,44,48,49,55,56,57] We have found that the judicious administration of sulfonamides, penicillin, and streptomycin makes possible the use of tantalum in contaminated and infected cases as well.

In this series of 106 consecutive tantalum cranioplasties the implants have all been fashioned by cutting and hammering the tantalum sheet into the desired shape during the operation. Excellent shaping may be accomplished using a ball hammer and a concave wooden block. With care, more complex contours, such as supra-orbital ridges, may be duplicated accurately (Figs. 1, 2, and 3). None of the methods of fashioning implants before operation[1,29,30,34,47,59] has been employed. Only one complication has arisen as a result of faulty shaping of the implant (Case 47†). Almost all of the plates have been fastened tightly to the outer table of the skull with small tantalum screws. This is by far the best method of fixation. Accurate apposition of implant to bone is obtained by crimping down the edge of the implant, thereby preventing erosion of the overlying scalp by protruding edges.[36] The thickness of the sheets employed has been reduced in successive stages from .020 to .007 of an inch, the latter being of sufficient strength, more easily and accurately fashioned, and less expensive. Scalp incisions over the implants have been closed with a single buried tier of interrupted black silk sutures in the galea.

The 106 cases are classified in 4 groups: (1) old defects—26 cases, (2) fresh, operative defects—55 cases, (3) contaminated traumatic defects—12 cases, (4) infected, operative defects—13 cases.

REPAIR OF OLD DEFECTS

(26 Cases)

Grant and Norcross[25] have listed 5 indications for the closure of old defects of the skull.

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* Read at the 38th Meeting of the Society of Neurological Surgeons in Cleveland, June 7, 1947.

† These cases cover a period of 49 months from February, 1943, to March, 1947. They have been numbered in chronologic order from 1 to 106.

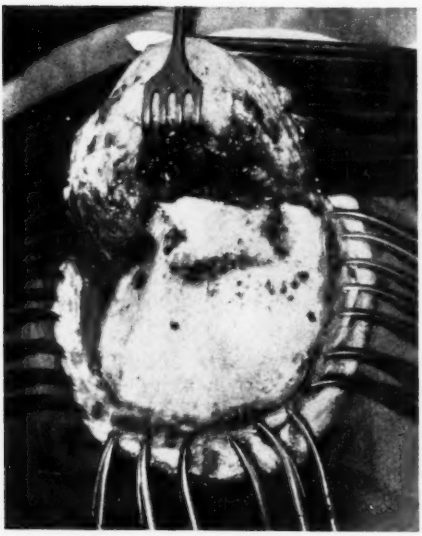

FIG. 1. Case 102. Frontal osteomyelitis involving supra-orbital ridge.

FIG. 2 .Case 102. Implant formed at operating table has replaced defect resulting from excision of infected bone.

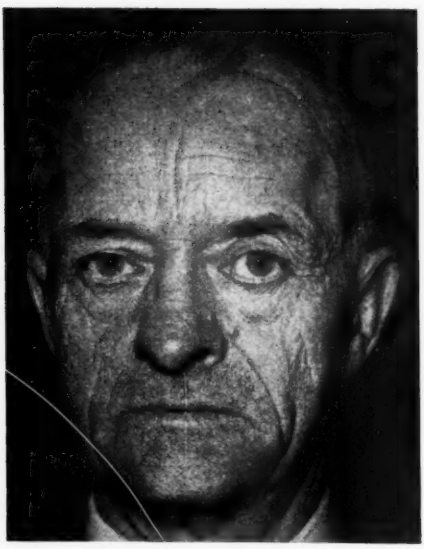

FIG. 3. Case 102. Cosmetic result. Retraction of the upper lid is due to scar of previous draining sinus.

1. Syndrome of the trephined.
2. Epilepsy resulting from the injury causing the defect.
3. Protection of the brain from possible trauma.
4. Unsightly defects.
5. Painful or unduly pulsating defects.

Mayfield and Levitch[36] have deleted traumatic epilepsy from this list of indications, feeling that closure of the defect has no beneficial effect. Gardner[19,20] has stressed the importance of closing pulsating defects, believing that the resulting reestablishment of the normal, non-pulsating state

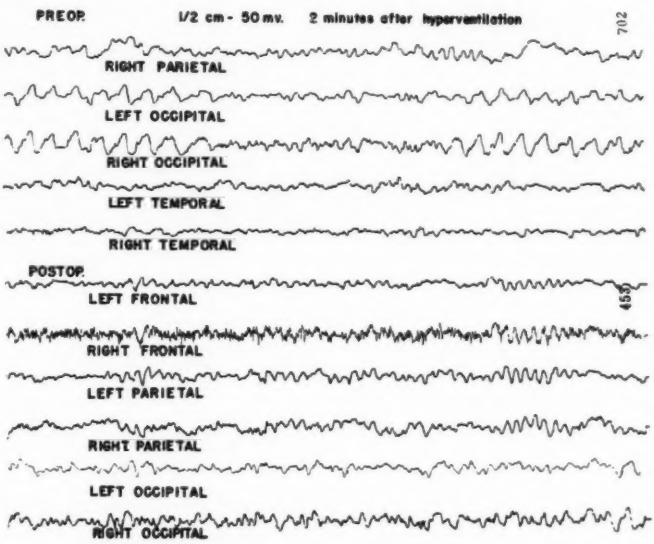

FIG. 4. Case 86. EEG before and after tantalum repair of cranial defect in right occipital area.

of the brain will reduce morbidity by diminishing progressive gliosis and atrophy beneath the defect.

In the group of 26 patients with old cranial defects repaired by tantalum implants there were 10 cases of traumatic or postoperative epilepsy, 6 cases with the syndrome of the trephined, and 10 cases of cosmetic repair. Of the 10 patients with traumatic epilepsy (Table 1) 6 have been completely relieved of seizures, 1 for 48 months, 1 for 36 months, 2 for 30 months, 1 for 24 months, and 1 for 10 months. In 3 cases the convulsions have been reduced in frequency. In 1 patient the epilepsy has not been materially changed. Comparison between preoperative and postoperative electroencephalograms in 1 of these patients (Case 86, Fig. 4) revealed uniform re-

TABLE 1

Traumatic epilepsy with skull defects

Case No.	Age Sex	Nature of Trauma	Duration of Defect	Seizures		Average Frequency	Operation Performed	Results
				Type	Duration			
12	M 20	Frontal craniectomy for osteomyelitis	22 mo.	Grand mal	22 mo.	1 per mo.	Cranioplasty	No attacks in 4 yrs.
4	M 43	Posttraumatic effusion and subtemporal decompression	15 yrs.	Akinetic	4 yrs.	1 per mo.	Cranioplasty	No attacks in 2 yrs.
17	M 45	Removal meningioma with decompression	16 mo.	Jacksonian	16 mo.	1 per wk.	Cranioplasty	No attacks in 2½ yrs.
29	M 44	Compound skull fracture with debridement	15 mo.	Grand mal	6 mo.	3 per wk.	Cranioplasty	No attacks in 2½ yrs.
30	M 12	Skull fracture at 7 mo.	11½ yrs.	Grand mal	2 yrs.	1 per wk.	Excision cerebral scar. Cranioplasty	No attacks in 3 yrs.
86	F 19	Skull fracture at 9 mo.	18 yrs.	Jacksonian	3 yrs.	2 per mo.	Cranioplasty	No attacks in 10 mo.
38	M 40	Compound skull fracture with debridement	2½ yrs.	Grand mal	2½ yrs.	3 per wk.	Excision cerebral scar. Cranioplasty	1 sensory Jacksonian per wk. for 3 yrs.
44	M 18	Compound skull fracture with debridement	4 yrs.	Grand mal	3 yrs.	Several daily	Excision cerebral scar. Cranioplasty	1st yr. 4 to 6 attacks per mo. 2nd and 3rd yrs. 1 attack per mo.
52	M 35	Removal meningioma with decompression	3 yrs.	Jacksonian	1 yr.	1 per wk.	Cranioplasty	2 attacks per mo. for 6 mo.
2	F 45	Compound skull fracture with debridement	20 yrs.	Grand mal	19 yrs.	1 to 2 per week	Cranioplasty	No change in 6 mo.

duction of dysrhythmia, less delta activity, lower voltage potentials from the focal area, and increased alpha activity.

The following history is particularly striking.

Case 12. In August, 1941, a previously healthy young man of 18 developed acute pansinusitis complicated by left frontal osteomyelitis, subperiosteal and extradural

abscesses. The surgical treatment included radical removal of the involved bone, followed by repeated incision and drainage of recurrent extradural abscesses. Convalescence was stormy. Complete recovery resulted, and the patient was discharged November 15 with the skull defect unrepaired. By the following August he had experienced 5 grand mal convulsions, and, in spite of dilantin and phenobarbital administration, the attacks became more and more frequent. In May, 1943, he was admitted in *status epilepticus* of 8 hours' duration. After the convulsive state was controlled, a tantalum cranioplasty was performed. The dura, found tightly adherent to the scalp, was separated by sharp dissection. The bony defect, measuring 7 by 9 cm., was closed without intradural exploration. Healing was uneventful. The patient was discharged with instructions to continue the dilantin and phenobarbital as before. He was last seen in April, 1947, 4 years after cranioplasty. He has not had a convulsion since operation and has taken no medication for the past 2 years.

In the group of cases of old defects, 6 patients exhibited the syndrome of the trephined, with varying degrees of headache, dizziness, tinnitus, fatigue, and inability to concentrate. Of these, 4 were completely relieved by tantalum cranioplasty, 1 improved, and 1 required subsequent ligation of the middle meningeal artery before complete relief was obtained.

One patient in this group presents an especially gratifying result.

Case 4. In 1930 a previously healthy young man of 30 was seen 2 weeks after being struck a glancing blow on the vertex by a heavy wooden beam falling 16 feet. Unconsciousness was momentary, and there was no residual paralysis. Symptoms consisted of generalized headache and vomiting. Examination revealed no physical or neurologic findings other than bilateral papilledema of 1 to 2 D. The spinal fluid pressure was 280 mm. of water and examination of the fluid revealed normal findings. Pneumoencephalography disclosed normal ventricles with poor delineation of the sulci. In May, 1930, a right subtemporal decompression was performed, with drainage of a subdural effusion, followed in October and again in February, 1931, by operations for the drainage of recurrent subdural fluid collections. The right subtemporal decompression remained tense and bulging for several months. The tension then subsided, but a pulsating hernia remained at the site of the decompression. He was completely disabled because of headache, dizziness, constant fatigue, and auditory hallucinations sometimes associated with akinetic seizures. In February, 1945, after 15 years of futile efforts to give him relief it finally occurred to us that these symptoms were probably due to the cranial defect rather than to the original injury. An encephalogram at this time revealed dilatation and wandering of the right lateral ventricle toward the decompression. A tantalum cranioplasty was performed, closing the 8 by 10 cm. subtemporal decompression. Healing was uneventful, and there was complete relief of symptoms. The patient was last seen 2 years after closure of the defect, completely asymptomatic and working every day.

Ten patients in the group with old defects presented large, unsightly, pulsating, and occasionally painful defects which were closed with tantalum implants without incident. One case in this group has been reported in detail elsewhere.[22]

In this group of 26 cases there were 4 postoperative infections, 1 resulting in death from a Staphylococcus aureus meningitis (Case 21, previously reported[20]). This death must be attributed to an error in judgment in oper-

ating too soon after a scalp infection. Penicillin was not available at that time. The other 3 infections were successfully treated with sulfadiazine and penicillin without drainage of the infection or removal of the implants.

Two patients in the group required removal of the implants.

The first (*Case 27*) had had a cranioplasty 4 months after the removal of a sequestrated bone flap. A thin scarred area was present in the scalp at the center of the defect, the site of previous drainage. In the preparation for cranioplasty this thin avascular spot was inadvertently "button-holed" and was closed with two sutures. Four days postoperatively a small fistula appeared, healed, then broke down again 2 months later. Temporizing measures failed to close the widening area of exposed tantalum, and the implant was removed 3 months after insertion. In retrospect, it is obvious that the thin avascular scar should have been revised before considering cranioplasty.

The second patient requiring removal of the implant (*Case 47*) had received a compound, comminuted fracture necessitating removal of a portion of the frontal bone, supra-orbital ridges, both frontal sinuses, and portions of the nasal bones, crista galli, and cribriform plate. Cranioplasty was performed 1 month after the injury. Four months after closure of the defect a sharp edge of the tantalum implant, replacing the left supra-orbital ridge, broke through the skin. Temporizing measures again failed, and the implant was removed 6 months after cranioplasty. The loss of this implant was due to the technical error of leaving a sharp rather than a rounded edge of tantalum to replace the supra-orbital ridge.

In neither of these cases was there meningeal or intracerebral extension of the infection surrounding the exposed plates, which is the complication feared by Bradford and Livingston.[3] On the other hand, a certain advantage resulted, even though the implants were lost, because the granulation tissue surrounding the implant, which these authors refer to as "unhealthy," caused enough connective-tissue thickening to bridge the bony defect effectively.

REPAIR OF FRESH OPERATIVE DEFECTS

(*55 Cases*)

Under this heading we have placed 24 cases in which closure with tantalum was performed when it was necessary to sacrifice portions of the skull involved by tumor, 18 cases in which both the free bone graft and a tantalum implant were used, 5 cases in which the craniotomy exposure was obtained with rongeurs, and 8 cases of controlled decompression. Of the first group, 14 patients had meningiomas, 3 had metastatic carcinomas, 3 had cholesteatomas, 2 had hemangiomas, and 1 each a periosteal fibroma and an osteoid osteoma. When the dura was excised with the involved bone no attempt was made to close the dural defect. Primary healing occurred in all cases, although there were 3 postoperative infections, one of which was a Streptococcus faecalis meningitis. The infections subsided on intensive antibiotic therapy without drainage or removal of any of the implants. Two months after primary healing 1 other patient (Case 35) developed necrosis at the center of the scalp flap where it had been invaded by tumor. There was no evidence

of infection until after the thinned avascular area had become necrotic. At reoperation for removal of the implant there was a thick layer of granulation tissue above and below the implant. Healing was uneventful.

In 8 cases both the free bone graft of the craniotomy* and a tantalum implant were used to close the operative defect. In these cases the craniotomy exposure proved inadequate for the removal of the brain tumor so that additional bone was rongeured away. The defect was then closed, using the free bone graft and in addition a tantalum implant (Figs. 5 and 6). Seven

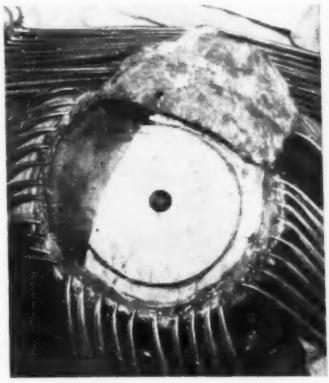

FIG. 5. Case 31. Showing closure of craniotomy defect by free bone graft and tantalum.

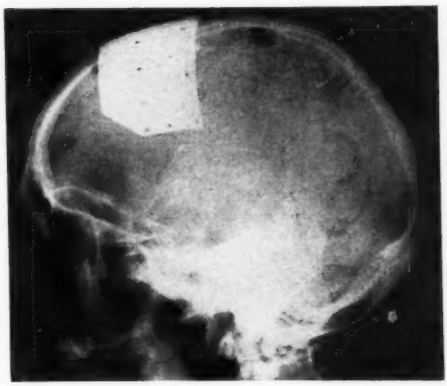

FIG. 6. Case 31. Postoperative roentgenogram.

had uncomplicated healing. The 8th case offers a striking contrast between the behavior of a free bone graft and a tantalum implant in the presence of infection.

Case 58. A left parasagittal meningioma was removed from a 55-year-old man after enlarging the original craniotomy opening. At this operation granulation tissue was found adjacent to the tumor, culture of which disclosed Staphylococcus aureus. The report of this culture was overlooked, and postoperative antibiotic therapy was not used. The operative defect was closed with the free bone graft and a crescent-shaped tantalum implant. Primary healing resulted, but the patient returned 2 months later because of pain and redness of the scalp flap. Aspiration revealed pus which grew Staph. aureus in pure culture. Penicillin (50,000 units) was injected beneath the scalp flap 3 times a day for 5 days, and the infection subsided. It reappeared 4 months thereafter with draining sinuses. At reoperation the bone disk was eroded, necrotic, and avascular, surrounded by thick granulation tissue. The tantalum implant, practically in contact with the infected bone disk, was surrounded by a thin, smooth membrane with no granulations or other evidence of infection. It

* At the Cleveland Clinic, in the usual craniotomy, a circular section of the skull is removed with a craniotome and is subsequently replaced as a free graft.

appeared that the bone disk acted as a reservoir for the infection, which, however, did not spread to the neighboring tantalum implant. The bone disk and tantalum implant were removed and healing was uneventful. The cause of the original infection adjacent to the tumor remains unexplained.

In a series of 10 cases of Jacksonian epilepsy the inner table of the removed bone flap, before being replaced, was lined with a thin sheet of tantalum in order to determine whether the metal itself had any anticonvulsant property. This treatment was employed in these cases since the lesions, because of their nature or location, did not lend themselves readily to ex-

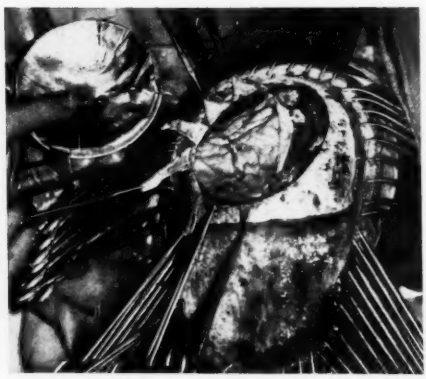

FIG. 7. Case 92. Illustrating method of replacing dura with sheet of tantalum fastened to the bone.

FIG. 8. Case 92. The bone with attached tantalum has been replaced.

cision. In 4 cases the convulsions were due to porencephalic cysts, in 2 to posttraumatic meningocortical adhesions, in 1 to a generalized gliosis in a retired prize fighter, and in 1 to a slow-growing glioma in Broca's area. In 2 patients no lesion was identified. In all cases the site selected for craniotomy coincided with electroencephalographic as well as clinical localization. In only the 2 cases of meningocortical adhesions was manipulation of the lesion done; in these cases the dural adhesions were gently separated from the cortex.

In all 10 cases the dura was excised over the exposed cortex and the cut edges everted over the rim of the craniotomy opening. A sheet of tantalum .004 inch thick was molded to the inner surface of the free bone graft and fastened with a tantalum screw. Four elongated tabs of the implant were then bent back, to emerge on the outer surface of the skull after the bone was replaced. This technic (Figs. 7 and 8) of placing a thin sheet of tantalum in contact with an epileptogenous focus was predicated on 3 reasons. First, in our series of cases of traumatic epilepsy, simple tantalum closure of the overlying skull defects appeared to benefit the seizures. We interpreted

these results as perhaps due to a grounding effect of the metal in carrying off the electrical charges of the incipient attacks before they reached convulsive levels. Second, we observed that in 23 patients with tantalum implants in which postoperative electroencephalograms were made, there appeared a uniform reduction in voltage potential over the site of the implant. Third, the procedure appeared innocuous since it has been shown[9,41,44] that tantalum foil placed in the subdural space prevents meningocerebral adhesions and is encapsulated in a thin diaphanous membrane without arachnoid thickening and with preservation of the intact cerebrospinal fluid pathways.

The dura was excised in order to bring the metal into as close contact as possible with the discharging focus. Electroencephalograms were made both preoperatively and postoperatively in 8 of the 10 patients. After operation there was uniform reduction in the voltage potential over the area of the implants, but in no case was there replacement of the abnormal delta waves and slow frequency with normal alpha activity. We believe that the reduced voltage potential is due to electrical interference by the implant and not to a reduction of the electrical activity of the cortex.

The results of this method have been poor. Of the 10 patients thus treated, 1 has been slightly improved, 8 have remained unchanged, and 1 has had more frequent and severe attacks. In no case has there been any complication arising from the metal-lined bone graft.

In 5 cases of recurrent brain tumor the operation was expedited by making the bony opening with a rongeur instead of attempting to elevate the bone graft of the previous operation. The defect was then closed with tantalum. All healed primarily, and no implants were removed.

During operations for unremovable gliomas when herniation of the brain has prevented replacement of the bone disk we have at times substituted a tantalum implant. This procedure has been employed in 8 cases, the bony defects being closed with implants fastened to the outer table and having the maximum convexity over which the scalp could be approximated. This procedure increases the intracranial space by at least the volume of the discarded bone. It restrains the decompressed brain and thus lessens edema and preserves functioning tissue, while obviating the unsightly cerebral hernias so frequently seen in terminal phases of gliomas. Primary union resulted in 6 cases. Two patients operated upon *in extremis* died within 72 hours. We believe that the surviving patients lived more happily until the tumors had filled the added space, after which they died without passing through the prolonged vegetative state common to similar cases with large decompressions. The only disadvantage of the method is that it hampers the radiologist in those cases in which x-ray treatment is indicated.

In this group of 55 cases the frontal sinuses were opened, with penetration of the mucous membrane, in 3 instances. A graft of cranial periosteum was placed over the rent in the membrane in each case, and no complications ensued.

REPAIR OF TRAUMATIC, CONTAMINATED DEFECTS
(*12 Cases*)

Twelve patients with craniocerebral penetration (6 gunshot wounds and 6 compound fractures) have received tantalum implants at the time of primary debridement. Four reached surgery within 8 hours, 4 within 16

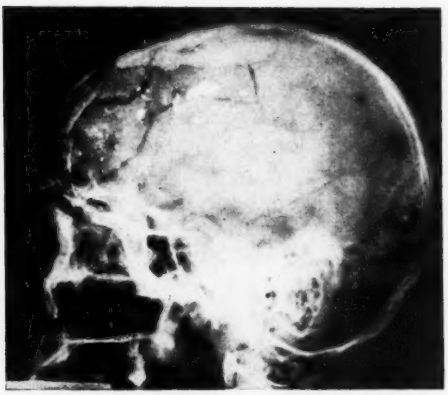

FIG. 9. Case 89. Gunshot wound, left frontal lobe.

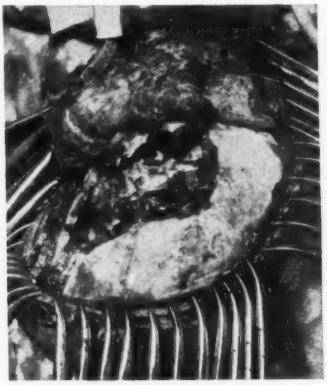

FIG. 10. Case 89. Appearance of wound during debridement.

FIG. 11. Case 89. Primary tantalum closure of contaminated wound.

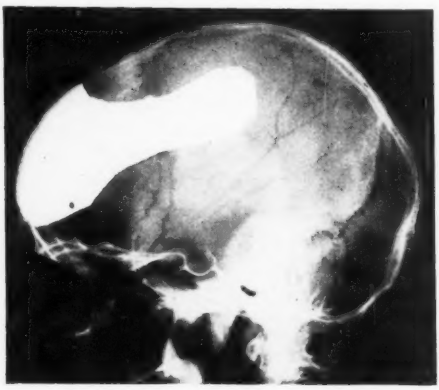

FIG. 12. Case 89. Postoperative roentgenogram.

hours, and the remaining 4 in 4, 6, 7, and 9 days. The operative treatment (Figs. 9, 10, 11, and 12) included as thorough debridement of bony and other fragments as was feasible, removal of blood clots and macerated brain by gentle irrigation and suction, then careful hemostasis. Sulfanilamide was

dusted into the wounds. The dura was closed tightly in 3 cases, partially in 6, and left unclosed in 3. Postoperative treatment included intensive administration of penicillin and sulfadiazine, and injections of 25,000 units of penicillin through the scalp down to the implant twice daily for several days. Primary healing occurred in all cases. These patients have been followed for from 7 to 48 months, the average being 23 months. Epilepsy has developed in 2 cases, or 17 per cent, which is the percentage reported by Maltby[35] in a series of 200 craniocerebral war wounds.

In 5 of these cases the cranial defect included the frontal sinuses. Free periosteal grafts were placed over the mucous membrane lacerations in 3 cases, and all healed without incident. In the 4th case it was necessary to remove the upper half of each frontal sinus after which the mucous membrane remaining was thoroughly removed and the nasofrontal ducts were plugged with pieces of gelfoam dipped in thrombin solution. This patient also healed without incident. In the 5th case cerebrospinal rhinorrhea and an intracranial pneumatocele developed 1 month after debridement and cranioplasty.

This patient was a 15-year-old boy (*Case 67*) who had been shot through the right frontal sinus, the bullet emerging to the left of the midline near the coronal suture. The upper portion of the frontal sinus and adjacent frontal bone were replaced by a tantalum implant at the 1st operation. The dura was closed but the mucous membrane was not. Primary healing resulted but the patient returned 1 month after operation with cerebrospinal rhinorrhea and an intracranial pneumatocele. At reoperation a minute perforation in the mucous membrane of the sinus was found and closed with a periosteal graft. The implant was then replaced. Primary union again resulted, and there was no recurrence of rhinorrhea or pneumatocele.

This case closely parallels the 2 reported by Woodhall and Cramer.[58]

Tantalum cranioplasty at the time of primary debridement has been advocated by Fulcher,[16] Echols and Colclough,[12] Robertson,[43] and Gardner,[19] the latter stressing that immediate repair of the defect immobilizes the wounded brain and scalp, thereby accelerating healing. In addition we believe, at least with conditions as they prevail in civilian practice, that tantalum closure can best be accomplished before the traumatized extracranial tissues have become cicatrized. As a result of their experience with fresh war wounds, Woodhall and Spurling[59] concluded that primary tantalum closure should be employed only in those cases without dural penetration. We believe that the procedure is even more important when dural penetration has occurred, since the interposition of the plate prevents adhesions between the extracranial and intracranial structures. If the dura overlying the traumatized cortex is excised as well, adhesions between the cortex and meninges may be prevented.

We are inclined to part with tradition in regard to the necessity of dural closure in these cases. We believe that when the skull is closed *with a tantalum implant*, closure of the underlying dura accomplishes one purpose. It helps to diminish the escape of cerebrospinal fluid into the extracranial

tissues during the postoperative period. The use of fascial or periosteal grafts or sheets of other organic material for closure of the dura affords the neurosurgeon the satisfaction of knowing that he has restored the structures to as nearly normal a state as possible but probably does not reduce morbidity. It renders the re-elevation of an osteoplastic flap a less traumatizing procedure, but it in no way aids the re-exposure of an area covered by a tantalum implant.

REPAIR OF INFECTED, OPERATIVE DEFECTS

(*13 Cases*)

Tantalum has been used in the primary repair of skull defects in 13 cases of infection (Table 2). Four of these patients had acute frontal osteomyelitis secondary to sinusitis, 1 had sequestration of the bone flap of a previous operation for brain tumor, and 8 had cerebral abscess. Two of the patients with osteomyelitis also had cerebral abscesses, making a total of 10. One of these abscesses was treated by cannula drainage and penicillin irrigation without excision of the abscess. This patient subsequently developed convulsions. The remaining 9 were treated by complete excision of the abscess, and, of the 7 who survived, 6 had no seizures and the 7th has had a single attack.

TABLE 2

Tantalum implants in infected cases

Case No.	Sex Age	Diagnosis	Surgical Procedure	Culture	Wound Healing	Results
34	M 17	Lt. parietooccipital abscess; frontal sinusitis	Total excision	Str. anaerobius	Per primam	Alive, well 39 mo. postop.
49	F 33	Rt. temporal abscess; otitic	Total excision	Pneumococcus Type I	Per primam	Alive, well 31 mo. postop.
50	M 6	Rt. frontal abscess from orbital cellulitis	Total excision	Str. salivarius	Per primam	Died 39 days postop. Recurrent brain abscess
56	M 49	Lt. temporal abscess; chronic mastoiditis	Total excision	Str. anaerobius P. vulgaris	Infection. Tantalum removed on 30th postop. day	Alive, well 28 mo. postop.
61	M 15	Rt. temporal abscess; chronic mastoiditis	Total excision	B. coli Staph. albus	Infection. Tantalum removed on 50th postop. day	Alive, well 25 mo. postop.
59	M 36	Rt. parietal abscess; metastatic from pneumonia	Total excision	Str. mitior	Per primam	Died 3½ mo. postop. Concomitant temporal and occipital abscesses

TABLE 2—*continued*

Case No.	Sex Age	Diagnosis	Surgical Procedure	Culture	Wound Healing	Results
65	F 23	Lt. occipital abscess; metastatic from tuberculosis of tibia	Total excision	Tubercle bacillus	Per primam	Alive, well 23 mo. postop.
91	M 8	Lt. frontal abscess; frontal sinusitis	Total excision	No growth	Per primam	Alive, well 9 mo. postop.
55	M 17	Frontal osteomyelitis. Lt. frontal abscess	Craniectomy Cannula drainage of abscess	A. faecalis Str. faecalis	Per primam	Alive, convulsions, otherwise well, 28 mo. postop.
82	M 22	Frontal osteomyelitis. Multiple lt. frontal abscesses	Craniectomy Total excision	Staph. aureus	Per primam	Alive, well 12 mo. postop. 1 convulsion
88	F 14	Frontal osteomyelitis	Craniectomy	Non-hemolytic Streptococcus	Per primam	Alive, well 7 mo. postop.
102	M 55	Frontal osteomyelitis. Draining subperiosteal abscess	Craniectomy Sinus closed	B. pyocyaneus Staph. albus Non-hemolytic Streptococcus A. faecalis	Per primam. Recurrent infection cleared	Alive, well 5 mo. postop.
90	F 41	Postop. wound infection. Brain tumor	Removal of sequestrum	No growth (48-hr. delay in culture)	Per primam	Died 60 days postop. Recurrent tumor

Four additional patients have been operated upon since these data were compiled. Two with intracranial abscesses and 2 with acute frontal osteomyelitis had primary tantalum cranioplasty at the time of the original operation and healed without complications.

Bradford and Livingston[3] believe that radical removal of a brain abscess is a questionable procedure. However, there are others who feel that complete excision of the abscess in conjunction with intensive administration of antibiotics is the rational treatment. The work of Bucy,[4] Furlow,[18] Rowe,[45] and Walker,[54] shows the efficacy of sulfonamides and penicillin in the adjuvant treatment. The reports of Vincent,[52] Groff and Grant,[26] Sachs,[46] Fincher,[15] and LeBeau[33] leave little room for doubt that total excision results in a lower mortality, less frequent recurrence, and lessened morbidity than more conservative methods.

A deterrent to radical excision, however, has been postoperative cerebral fungus and sequestration of the bone flap. Closure with tantalum prevents these complications and does not increase the possibility of postoperative meningitis. This last point is demonstrated by 2 cases of temporal lobe

abscess associated with chronic mastoiditis. Treatment consisted of tota excision of the abscess and tantalum closure of the skull defect. The infectior in the adjacent mastoid spread to the implants. Although the dura was left widely open beneath the implants in these cases, meningitis did not develop. The implants were removed after several weeks, and the tissue thickening resulting from the infection rendered further cranioplasty unnecessary. Both patients are alive and well 25 and 28 months after operation.

There is considerable experimental[20] and clinical evidence[6,7,21,23,28,58] that tantalum does not deter healing in infected wounds. This is corroborated by the 4 cases of frontal osteomyelitis in this series (Table 2) in which tantalum implants were used to close the defects at the time of the removal of infected bone. The implants were retained, and the cosmetic results were excellent. The results obtained were made possible by intensive parenteral administration of penicillin and oral administration of sulfadiazine as well as local injections of penicillin. In addition, the last 3 patients received urea, as advocated by Ecker[13] and LaLonde and Gardner,[31] to enhance the action of the sulfonamide. Primary wound healing occurred in all 4. The last case offers particularly convincing evidence of the inertness of a tantalum implant in the presence of infection.

Case 102. A 55-year-old man was admitted with left frontal osteomyelitis arising from an acute sinusitis (Figs. 1, 2 and 3). There was a draining sinus beneath the supra-orbital ridge resulting from the incision of a subperiosteal abscess. Culture of the pus revealed a mixed infection of Bacillus pyocyaneus, Alcaligenes faecalis, Staphylococcus albus, and non-hemolytic Streptococcus. At operation a sheet of tantalum was cut and molded to the shape of the involved bone, which included the left supra-orbital ridge. After removal of the infected bone the implant was fastened in place with 3 tantalum screws and the draining sinus was closed with a tantalum wire suture. The scalp incision was closed without drainage, after which 25,000 units of penicillin were injected down to the implant. The postoperative antibiotic administration was as outlined above. Primary union resulted, but the patient returned 1 month later with a recurrence of drainage from the sinus beneath the supra-orbital ridge. Culture again revealed the same four organisms. Treatment consisted of administration of penicillin intramuscularly, sulfadiazine and urea orally, as well as alternate 2-hour injections of penicillin and streptomycin down to the implant through a ureteral catheter placed in a stab incision. Once daily the wound was barbotaged with 10 cc. of a 1 per cent acetic acid solution to combat the Bacillus pyocyaneus component of the infection. The draining sinus was again closed with a tantalum wire suture. The infection subsided without removal of the implant, and there has been no further recurrence.

As can be seen (Fig. 3), the cosmetic result is excellent. It represents the only case in the series in which tantalum was implanted in a *draining* wound. Prior to our experience with this case we had not believed it possible to obtain primary healing under these circumstances because of the presence of secondary invading organisms.

In this group of 13 infected, operative defects with primary tantalum

closure there were 3 deaths. One death, early in the series (Case 50), was due to a recurrent abscess at the primary site. This recurrence was due to the use of too small and too few doses of penicillin. The passivity of the implant in this case was indicated by the fact that the recurrent infection did not involve it. The 2nd death (Case 59) was due to associated metastatic

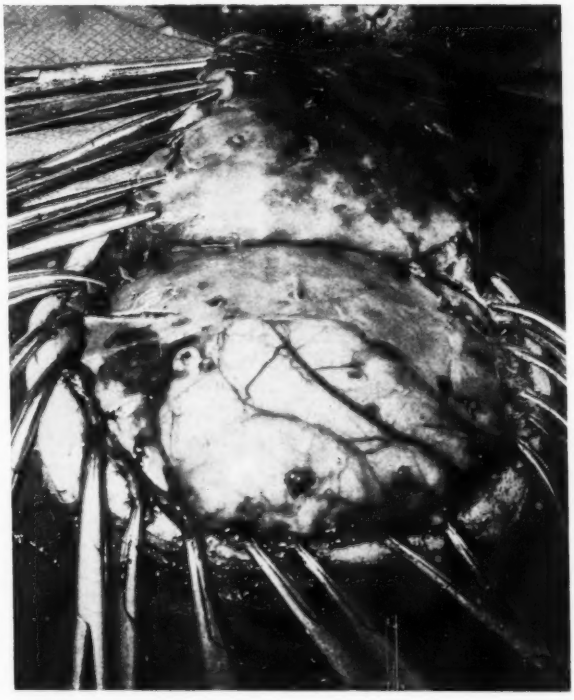

FIG. 13. Case 59. Appearance upon removal of tantalum implant 50 days after insertion. Convolutions of brain plainly visible through area of newly-formed dura.

abscesses in remote portions of the brain which could not be located. The 3rd death was due to regrowth of a brain tumor.

This patient (*Case 90*) had an operation for a glioma of the left temporal lobe. She returned 24 days after the craniotomy with meningitis and cerebral hernia. At reoperation the bone plate was found to be infected. After removal of the bone and evacuation of the underlying abscess, the cranial defect was closed with a tantalum implant. The infection subsided on antibiotic therapy and the wound healed by primary union. Sixty days later the patient died from regrowth of the glioma. There was no recurrence of infection.

In these cases of infection, the usual postoperative antibiotic treatment

consisted of 50,000 units of penicillin intramuscularly every 3 hours and 25,000 units injected through the scalp down to the tantalum implant every 12 hours for 3 days, then once daily for another 4 days. In addition, sulfadiazine was administered orally in amounts sufficient to produce a blood level of 10 mg. per cent. Some patients were given urea by mouth in order to enhance the effectiveness of the sulfadiazine. The usual dose of urea was 30 gm. every 4 hours but varied somewhat to maintain a blood level approaching 100 mg. per cent.

FLUID ACCUMULATION OVER THE IMPLANT

The dura was excised or left unclosed in 58 per cent of the 106 cases in this series. Troublesome cerebrospinal fluid accumulation about the implant (arbitrarily defined as accumulations requiring 3 or more aspirations of 20 cc. or more), occurred in 16 cases. Further analysis reveals that five factors were probably responsible: (1) non-closure of the dura, (2) poor apposition of the edges of the implant to the skull, (3) inadequate convexity of the implant contour, allowing the redundant scalp to be lifted away from the tantalum, (4) direct communication between ventricular system and the implant, (5) perforations in the implant. There has been no case in which troublesome fluid formed with but one of these factors present, only 2 cases in which two factors were present, and the remaining 13 cases were associated with three or more of these factors. When this situation develops it can usually be corrected by aspiration of the fluid under rigidly aseptic conditions and firm application of elastic bandages to the area involved. With careful attention to the avoidance of the other four causes, we find that excision of the dura over the traumatized brain is not followed by troublesome accumulation of fluid over the implant, and it undoubtedly aids in the prevention of meningocortical adhesions.

The fluid that collects over the implant is not due to irritation or foreign body reaction. It consists of a mixture of blood and cerebrospinal fluid which escapes outside the skull and lifts the scalp away from the smooth outer surface of the implant. When the subarachnoid spaces become sealed off, the fluid no longer accumulates.

REOPERATION

Reoperation at the site of the tantalum implant has been performed in 12 cases from 1 to 15 months after cranioplasty. Two autopsies were performed 2 and 8 months after cranioplasty. In all cases the tantalum was as shiny and new-appearing as when inserted. In comparison to the difficult re-elevation of osteoplastic flaps, the elevation of the tantalum implants was simple, quick, entailed less blood loss, and resulted in no trauma to the cortex. The observations in these 14 cases do not support the statements of Pudenz[40] and Fulcher[16] that the scalp becomes adherent to the implant. Only at the perforations in the tantalum is the tissue adherent, and this is because of fibrous union between the two enclosing layers of the capsule. The im-

plant was encased uniformly in a thin membrane without evidence of exuberant fibrous overgrowth. In 4 cases in which the dura had been excised at the first operation there were no adhesions between the underlying cortical scar and the thin membrane beneath the implant (Fig. 13). This substantiates the observations of Cone, Pudenz, and Odom[9] and Robertson and Teacher,[44] in contradistinction to the findings of Delarue, Linell, and McKenzie.[11] The latter authors found serious dural thickening in 12 dogs after implanting tantalum foil in the subdural space.

SUMMARY

In this series of 106 consecutive tantalum cranioplasties carried out in civilian practice there have been 8 deaths, but only 1 can be attributed to the procedure *per se*. Eight implants have had to be removed, because of exposure of the implant in 4 instances, by a breaking down of the scalp from underlying infection in 1, because of infection in an adjacent mastoid in 2, and because of infection in an adjacent free bone graft in 1. None of the patients developed a secondary meningitis from the draining wound, a possible complication pointed out by Bradford and Livingston.[3] We believe that the closure of contaminated or infected cranial defects not only does not increase the likelihood of the development of meningitis but actually militates against it by preventing herniation and stopping the pulsation, and thereby aids healing. There has been perfect healing in all cases of compound fractures when the tantalum was inserted at the time of debridement and closure. A higher incidence of complications occurred when tantalum was used for secondary closure of old defects than in any other group. This suggests that in traumatic cases tantalum should be employed in primary closure rather than performing primary debridement alone with secondary repair of the defect at a subsequent operation.

The frontal sinuses were involved in the cranial defect in 13 of the 106 cases. Two of these were old defects, 2 were fresh operative defects, 5 were traumatic contaminated defects, and 4 were cases of frontal osteomyelitis. Only 1 of the 13 implants had to be removed. This was due to faulty shaping of the implant, which eroded through the skin. Lane and Webster[32] report 52 collected cases in which tantalum implants were removed. Of these, 35 per cent were cases in which the skull defect involved the frontal sinuses. These authors suggest that osteoperiosteal transplants may be used to greater advantage in this area. Our results are not in accord with this.

CONCLUSIONS

1. Traumatic epilepsy is usually benefited by closure of associated skull defects. This is not due to any property inherent in tantalum, since Jacksonian epilepsy without skull defect is not altered by implanting tantalum over the discharging focus on the cortex.

2. Tantalum cranioplasty may be used to provide a controlled decompression when cerebral herniation prevents replacement of the osteoplastic

flap. It enlarges the intracranial space and thus relieves pressure. It lowers morbidity and prohibits massive cerebral hernia in terminal stages of inoperable tumors. It does, however, make postoperative x-ray treatment more difficult.

3. Contaminated craniocerebral wounds with skull defects should be treated by tantalum closure at the time of the debridement.

4. It is not necessary, and at times not desirable, to close the dura beneath a tantalum implant.

5. With adequate antibiotic therapy, tantalum will heal in an infected field unless there is constant reinfection from a contiguous bony focus such as chronic mastoiditis.

6. The presence of a tantalum plate in a contaminated or infected wound does not increase the incidence of intracranial extension of the infection but actually helps to prevent it by immobilizing the wound.

7. When reoperation for brain tumor is anticipated, tantalum closure is preferable to replacement of bone because it renders the re-exposure a less traumatic procedure.

8. Complete coverage of the implant with healthy scalp is essential to successful cranioplasty.

9. The indications for tantalum cranioplasty may be summarized as follows:

a. Old defects
   1. Posttraumatic epilepsy with defect.
   2. Syndrome of the trephined.
   3. Large, unsightly, pulsating, or painful defects.

b. Fresh defects
   1. Involvement of skull by tumor.
   2. Extension of craniotomy opening to expose tumor.
   3. Exposure of recurrent tumor by rongeur.
   4. Discarding bone flap to provide controlled decompression.
   5. Discarding bone flap to facilitate later reoperation.
   6. Debridement of compound fracture.
   7. Excision of brain abscess.
   8. Excision of osteomyelitis of the skull.

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A STUDY OF 211 PATIENTS WITH VERIFIED GLIOBLASTOMA MULTIFORME

LOYAL DAVIS, M.D., JOHN MARTIN, M.D., STANTON L. GOLDSTEIN, M.D. AND MOSES ASHKENAZY, M.D.

Department of Surgery, Northwestern University Medical School, Chicago, Illinois

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. . . because I strongly believe that the operative mortality is higher in the case of gliomas treated merely by decompression than it is when the growth, once exposed, is removed as completely as possible, it is our invariable rule vigorously to attack a glioma of the cerebral hemisphere rather than to withdraw from it.—*Harvey Cushing.*

IN A follow-up study of 211 patients with glioblastoma multiforme, verified microscopically, we have found a relationship between the postoperative survival time and the pathological characteristics, the various types of surgical attack, the age of the patient, the site of the tumor, and roentgen

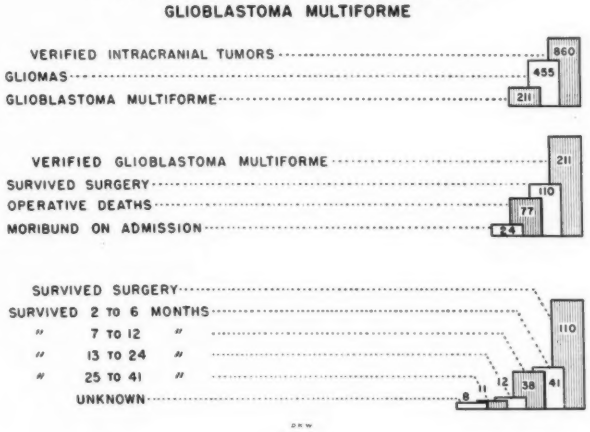

CHART 1

and radium irradiation. There also was a tendency for some of the tumors to change their histopathological character during treatment of the patients.

We have encountered 860 patients with verified intracranial tumors from July 1, 1924, to July 1, 1947 (Chart 1). Of these, 455 patients (52.9 per cent) had gliomas. The 211 patients who had verified glioblastoma multiforme comprised 29.5 per cent of the total number of intracranial tumors, or 46.4 per cent of the gliomas. Careful follow-up studies have been done in each instance, except for 8 patients in this group, whom we have been unable to follow.

Of the 211 patients, 24 were brought into the hospital in such poor condition that they expired shortly after arrival, before any surgical procedure was done. The diagnosis in each instance was verified by autopsy. The remaining 187 patients were operated upon 225 times, and of these, 77 died in the hospital within 1 month after operation. The cause of death in many cases was directly due to the operation, but any patient dying within 1 month after the operation, regardless of the cause, was considered an operative death. The case mortality was 36.5 per cent and the operative mortality was 30.2 per cent. Included in this number are 8 patients in whom ventriculography alone was performed, and who expired before any direct surgical attack upon the tumor was made. Of the 110 patients who survived surgery, 41 lived for 6 months or less, 38 survived for 7 to 12 months, 12 survived for 1 to 2 years, and 11 survived for longer than 2 years. The longest survival time was that of a patient who lived for 41 months after operation (Chart 1).

PATHOLOGY

We have based our microscopic diagnosis of glioblastoma multiforme on a histogenetic schema according to Bailey and Cushing's classification of gliomas. This histological division was not always clear-cut, because of the frequent cytological and structural variations encountered in different sections of the same tumor, particularly when only a small fragment of tissue was first available for biopsy at operation, and later, the whole tumor was restudied at postmortem examination. As a matter of fact, in addition to the 211 tumors studied, there were 14 which were originally classified as glioblastoma multiforme. However, when these tumors were critically and repeatedly re-examined, they were finally verified as astrocytomas (8), medulloblastomas (3), oligodendroglioma (1), spongioblastoma unipolare (1), and melanosarcoma (1).

Again, there was another group of 8 tumors in this series, 7 of which were verified as astrocytomas after operation, and 1 as an oligodendroglioma, which, after second operation or autopsy, showed a transition to glioblastoma multiforme (Figs. 1 and 2). These mixtures of different cytological glioma entities in one and the same tumor have been discussed previously by Carmichael,[5] Davidoff and Ferraro,[8] Cox,[6] and Deery.[10] These observers also confirmed Virchow's[26] original observations in 1863–65 and those of Stroebe,[27] Henneberg,[15] and Tooth,[25] on the transition of a so-called "benign" glioma into a more "malignant" type. Bailey and Cushing[1] and Globus and Strauss[12] also observed glioblastomas to dedifferentiate from astrocytomas.

In attempting to correlate the microscopic pathology of glioblastoma multiforme with the survival time of the patients, we were struck by the preponderance of proliferative vascular changes in these tumors, and, on the basis of their angio-architectural patterns, we arbitrarily divided them into two groups. The first, or *angiothrombotic group*, was characterized by microscopic evidence of thrombosis, both recent and organizing, with multiple large areas of infarction and hemorrhage (Fig. 3). The second, or

angioproliferative group, was characterized by adventitial or perivascular proliferations, glomerular formations or sessile bud-like outgrowths of the intima, or intravascular endothelial proliferations resembling at times even obliterative endarteritis (Gough[13]), but usually without any evidence of thrombosis at the site of subdivision (Fig. 4). If any thrombi at all were seen in these tumors, or any large areas of necrosis, as distinct from minor degenerative changes, then they were all placed in the angiothrombotic group regardless of the fact that co-existing extensive proliferative changes

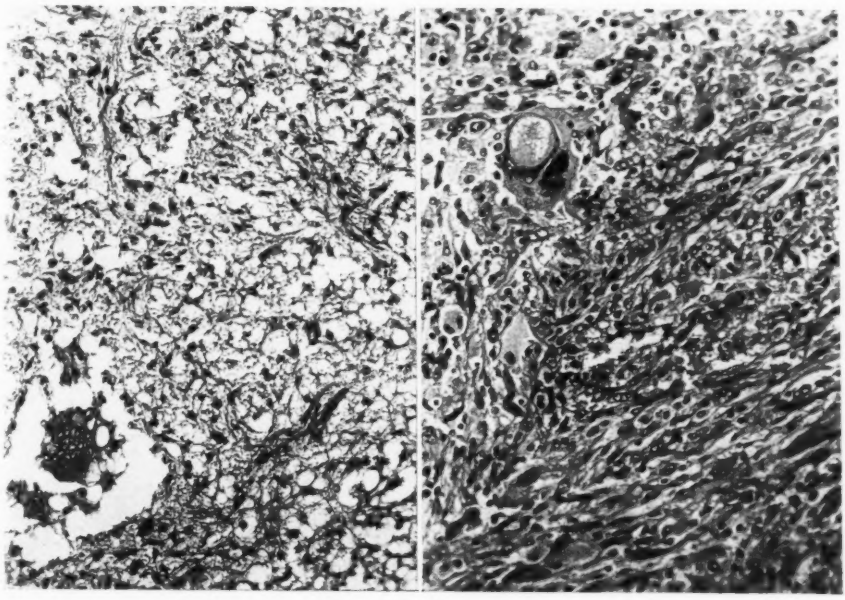

FIG. 1. (*left*). Typical astrocytoma, showing uniformity of cell types and some cyst formation. Compare with Fig. 2. (Hematoxylin-eosin, ×230.)

FIG. 2 (*right*). Tumor from same patient as in Fig. 1 seven months later, showing transition to glioblastoma multiforme. (Hematoxylin-eosin, ×230.)

may also have been present in the blood vessels. Despite this, there was still a group of 9 patients in which no clear-cut distinction could be made, and it was found impossible to classify the tumor on this angio-architectural basis.

There were 139 tumors designated as angiothrombotic, and 63 as angioproliferative. Of the 24 patients who were admitted in extremis and died without operation, 17 were in the angiothrombotic group, while 6 were in the angioproliferative group. Of the 77 patients who did not survive surgery, 60 were in the angiothrombotic group, and 14 were in the angioproliferative group (Chart 2). Of the patients who survived surgery, 10 (16.1 per cent) of

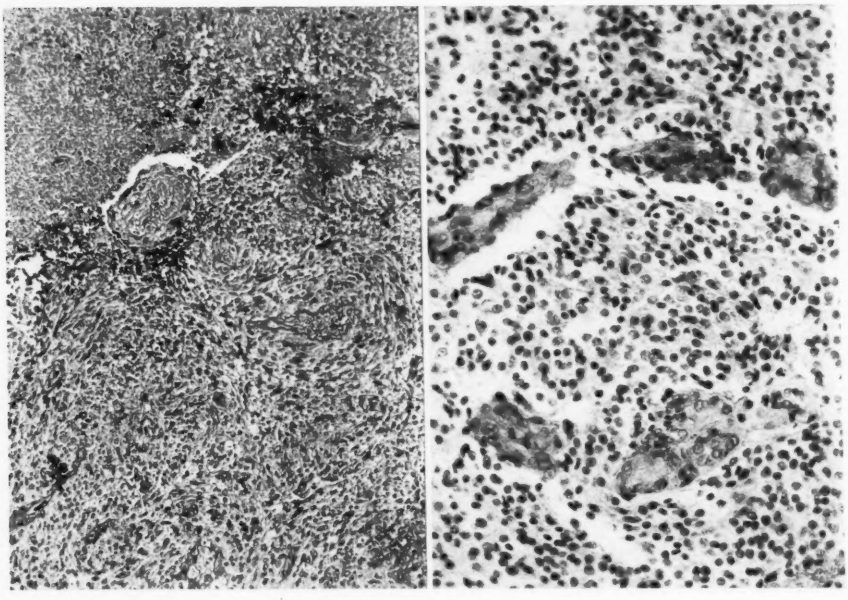

FIG. 3 (*left*). Angiothrombotic type of glioblastoma multiforme, showing thrombosis, hemorrhage and necrosis. (Hematoxylin-eosin, ×100.)

FIG. 4 (*right*). Angioproliferative type of glioblastoma multiforme, showing endothelial and adventitial proliferation of the blood vessels. (Hematoxylin-eosin, ×230.)

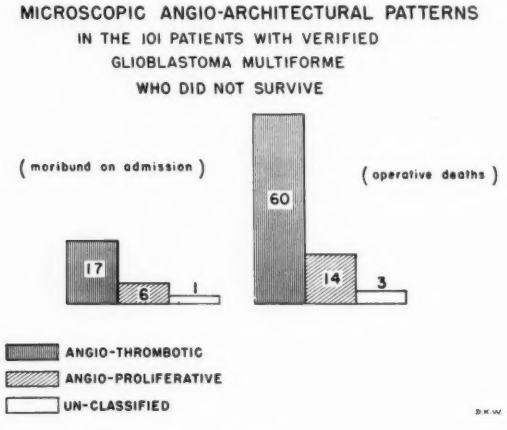

CHART 2

the angiothrombotic group survived for more than 1 year, while 13 (30.2 per cent) of the angioproliferative group survived for more than 1 year. Similarly 3 (4.8 per cent) of the angiothrombotic, and 8 (18.6 per cent) of the angioproliferative group survived for more than 2 years (Chart 3). These figures are statistically significant by the law of probable error and show that the angioproliferative group survived for a significantly longer period of time following surgery.

Tooth[25] in 1912 was the first to emphasize these vascular proliferative changes. He was able to subdivide the evolutionary process of an actively growing glioma into four stages, which he called angiomatous, intimal proliferative, thrombotic, and necrotic. Our angioproliferative group of tumors

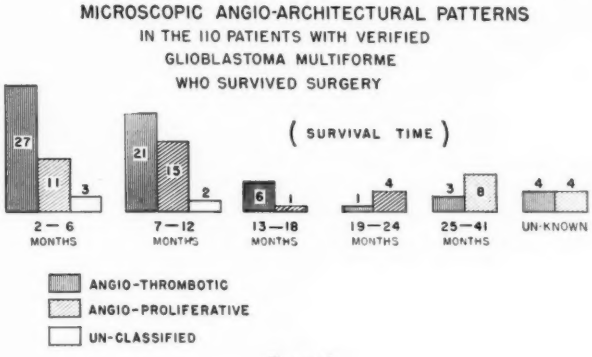

CHART 3

corresponded to his first two stages combined, and our angiothrombotic to his final two stages. Virchow[26] in 1865 and Stroebe[23] in 1895 noted these "telangiectatic" and hyperplastic blood vessels as part of the tumor process, and their significance was discussed later by Bertrand and Medakowitch,[3] Scherer,[20,21,22] and Bertha.[2] The vascular patterns and reactions of these tumors were also studied by Globus and Strauss,[12] Bailey and Cushing,[1] Carmichael,[5] Deery,[10] Elsberg and Hare,[11] and Hardman.[14] Sahs and Alexander[19] suggested that this awkward and inadequate vascular supply was a major factor in the causation of the frequent, widespread necrosis in glioblastoma multiforme.

Penfield[18] has been the strongest proponent of the view that this remarkable multiplication of endothelial cells would yet prove to be the essential part of the tumor, and this viewpoint seemed to be strengthened by the fact that at least the glomerular type of proliferation was rarely ever seen in any other tumor but glioblastoma multiforme. Bailey and Cushing,[1] Deery,[10] Gough,[13] and others considered these to be reactive blood vessel proliferations, perhaps on a toxic basis, since these formations in rare instances did appear in other glioma types as well, particularly in the astocytomas associated with dedifferentiation, and occasionally also outside the edges of a

metastatic carcinomatous nodule. Scherer[22] noted that these angioblastic proliferations have always been observed in patients with an extremely rapid evolution of signs and symptoms. This, together with the curious fact that they not only precede the growth of the tumor, but also grow from the uninvaded tissue towards the edge of the tumor, gave them a biological significance which he considered was worthy of further research.

In 1940, Scherer[22] studied the evolutionary stages and growth processes of the gliomas, and as a result divided his glioblastomas into a *primary* type, revealing extensive necrosis, and a *secondary* type, revealing comparatively small or no necrosis, usually with some small areas of cystic degeneration. From his description, we believe that these strongly resembled our angiothrombotic and angioproliferative groups respectively. His latter group were also those with "long clinical evolution," whereas his primary glioblastomas did not live beyond 6 to 12 months.

Busch and Christensen[4] subdivided their glioblastomas into three main pathological groups. These groups were: (1) *angionecrotic*, characterized by pleomorphism of cells, necrosis, hemorrhage and increased vascularity, with proliferation of intima and adventitia, as well as thrombosis, fibrosis, and hyaline degeneration; (2) *multicellular*, characterized by less vascularity, less polymorphism, and greater richness of cells; and (3) *magnocellular*, characterized by large, plump cells with eccentric nuclei and the more frequent occurrence of multinuclear giant cells. We attempted to classify our 211 glioblastoma multiforme tumors in this manner, but the attempt was only partially successful because in many cases there was no clear-cut differentiation between the types, and frequently there was merging of one type into the other in different sections of the same tumor. In our study, 96 tumors were angionecrotic, 68 multicellular, 38 magnocellular, and 9 unclassifiable. Of the patients who survived surgery, 8 (17. 0 per cent) of the angionecrotic lived for 1 year or longer, as contrasted with 11 (39.6 per cent) of the multicellular, and 4 (33.7 per cent) of the magnocellular group. This was in general agreement with the findings of Busch and Christensen,[4] but our results were not as dramatic as theirs.

AGE INCIDENCE

Over half of the patients were between 40 and 60 years old, the greater number being in the 40 to 50 year age group. However, the ages ranged from 3 years to 75 years, and there were 4 patients below the age of 10. Of the 4 patients in the 3 to 10 year group, it should be noted that none survived surgery, and the 2 over 70 years old were in extremis on admission and died without operation. There were 19 patients between the ages of 61 and 70 years, but none of these survived for more than a year. The patients in the third decade of life generally survived longer than the others. Of the 25 patients in the above three extreme age groups, 20 were classified as angiothrombotic. Again this seems to indicate that this group is the more malignant one.

SEX INCIDENCE

Several of the patients in this series were treated at a Veterans Administration Hospital, where there is a high preponderance of male patients. Therefore, we do not believe that our figures relative to sex incidence are representative.

LOCATION OF THE TUMOR

Of the 211 tumors, 103 were confined to either the frontal, parietal, or temporal lobe, most of them being in the latter location. In only 1 patient was the tumor confined to the occipital lobe. Seventy-eight tumors occupied more than one lobe but were unilateral, while 26 were bilaterally located in the cerebrum. Three tumors definitely verified as glioblastoma multiforme were infratentorial in location. One of these was located in the brain stem of a 51-year-old male and only a surgical biopsy was taken because of the inaccessibility of the tumor. He expired in the hospital shortly after operation and the tumor could not be classified definitely as angiothrombotic or angioproliferative. Another was located in the right cerebellar hemisphere, with extension into the pons, of a 12-year-old boy, who also died in the hospital shortly after operation. A subtotal resection of the tumor was done and the tumor was classified as angiothrombotic. The third tumor was located in the right cerebellar hemisphere of an 8-year-old boy, who died on the operating table during the induction of anesthesia, before the tumor was attacked. This tumor was angioproliferative.

Of the 30 tumors confined to the frontal lobe, 3 survived for more than 2 years, while of the 73 confined to either the temporal or parietal lobe, only 2 survived for more than 2 years. The operative mortality was only 27 per cent in the tumors confined to one lobe, while it was 44 per cent in the multilobar tumors, and 50 per cent in the bilaterally located tumors. Four patients with multilobar tumors lived for more than 2 years, while 2 with bilaterally located tumors lived for more than 2 years.

SURGICAL TREATMENT

It has been our invariable rule to operate through a large osteoplastic craniotomy in order to have adequate exposure of the tumor for removal and to provide, if necessary, a thorough decompressive effect. Even when there was good reason preoperatively to suspect the tumor of being glioblastoma, we have never followed the practice of performing a trephine opening to obtain a small amount of tissue by means of a ventricular needle or a small forceps. The histologic nature of any tumor should not be judged by such limited tissue study, and on several occasions we have found that the tissue so obtained and diagnosed elsewhere as "glioblastoma" or "probably glioblastoma" proved, on later removal of the tumor, to have been merely the softening with glial proliferation around a benign tumor or a glioma of much less malignant character than a glioblastoma. Neither do we feel that the discovery of cystic fluid in a softened area in the brain by such methods is sufficiently diagnostic of glioblastoma to deny the patient any benefit of

complete surgery. Were only such limited operative approaches persistently made to the surgical problem of the glioblastoma, the solution of this admittedly difficult situation would never be attained. The electrosurgical unit, improved instruments, adequate suction apparatus, and hemostatic agents have facilitated the radical removal of these tumors during the last 20 years.

Of the 187 patients who were operated upon, the tumor was not directly attacked in 39. These patients were those in whom there was an error in localization of the tumor, or on whom ventriculography was performed with death occurring before operation could be done, or those on whom subtemporal decompression alone was done. In the early years of our treatment of these tumors, and especially before the now common use of preoperative pneumoencephalography, subtemporal decompression was occasionally done as a first-stage operation, or to relieve the patient of his increased intracranial pressure while awaiting the appearance of further localizing signs or an improvement in his general condition. In the remaining 148 patients in whom the tumors were directly attacked, the involved lobe was completely resected in 7. Forty-one patients had resection of all grossly visible tumor and in many instances this amounted to over 100 grams of tissue. From 1 woman with a glioblastoma of the right frontal lobe 106.6 grams of tissue were removed at the first operation, and 90 grams more were removed at a second operation almost 2 years later. Seventy-one patients had subtotal removal of the tumor and 28 patients had smaller amounts of tumor removed for microscopic verification only when for special reasons a more radical operation was deemed unwise. Such partial removal of the tumors was done when the lesion lay adjacent to a vital area of the brain, when the tumor extended widely through the corpus callosum into the opposite hemisphere, or when the patient was in such poor condition that wide removal of the tumor could not have been tolerated.

As with tumors elsewhere in the body, it is a general rule that the wide removal of a glioblastoma undoubtedly prolongs the life of the patient. Of those patients who had complete removal of a lobe and survived surgery, 50 per cent lived for more than a year after operation. Twenty-six per cent of those who had resection of all grossly recognizable tumor, 21 per cent of those who had subtotal removal, and 18 per cent of those who had surgical biopsy only, survived for more than a year after surgery. Of the 39 patients in whom the tumor was not directly attacked, only 1 patient survived for more than a year.

The immediate postoperative state of the patient, as well as his survival time and his comfort and well-being during that period, is markedly affected by surgical decompression of the brain. At the time of operation the bone flap was removed in 90 patients, while the usual decompression in the flap was made in 80 others. There were 33 per cent operative deaths among those patients in whom the bone flap was removed, while in those in which the usual decompression in the bone flap was made the operative mortality was

45 per cent. The bone flap was always removed in those instances where the tumor was obviously of a highly malignant and infiltrative nature, or when the brain appeared to remain under tension even after resection of the tumor. The usual decompression in the bone flap was performed when the tumor appeared to be more benign in nature, when all visible tissue had been removed from a lesion that appeared to be fairly well circumscribed, or when, following removal of the tumor, the brain appeared to be relieved of its tension and not likely to undergo marked postoperative swelling. It is noteworthy that the postoperative mortality was actually lower in that group of patients with the most severe lesions and in whom the widest possible decompressive effect was obtained by removal of the bone flap. However, fewer patients in this group survived for more than a year after surgery than among those in whom a decompression in the bone flap was performed, indicating that with succeeding months the malignant and destructive characteristics of the tumors outweighed the immediate good effects of decompression. The approximate histologic nature of the tumor was verified by frozen section or by a rapid vital staining technique but these methods are not completely reliable.

The story of surgical accomplishment in this most difficult group of intracranial tumors is not a pleasing one. It should challenge the surgical conscience and ability of every neurosurgeon. New methods must be devised to supplant the present crude surgical techniques of glioma removal. The challenge cannot be met by adopting procedures which hasten rather than retard the clinical progression of the gliomas.

ROENTGEN IRRADIATION

Forty-three patients received roentgen irradiation and 15 received radium therapy. Of those who received more than 4,000 r units, 12 survived for more than 1 year, while of those who received less than 4,000 r units, none survived. Of those patients who survived surgery and received no irradiation, only 5 survived for more than 1 year. Only 3 of those who received radium irradiation survived for the same period, while the other 12 succumbed within the year. From our findings it would appear that those patients who received adequate roentgen irradiation therapy survived for a longer period of time than those who did not. However, despite these figures, it must be remembered that those who had inadequate roentgen irradiation therapy may not have lived long enough following surgery to either start, or complete, their courses, because of the relatively very malignant nature of their respective tumors.

We also studied the microscopic sections of 12 tumors both before and after roentgen irradiation therapy (Figs. 5 and 6), and our findings were as equivocal as those of other investigators[1,6,7,9,16,17,24] of this problem. As Deery[10] found, some of the glioblastomas showed striking histopathological changes, while others were less convincing, and still others showed no distinct changes at all. In 5 cases we found a striking increase in

the number of pyriform, undifferentiated, and giant cells, with marked bizarreness and hyperchromaticity of their nuclei. The stroma showed an increased number of extensive hemorrhages, and areas of necrosis and degeneration, with foci of replacement fibrosis, as well as of small round cell infiltration. The blood vessels in areas also showed more extensive changes, particularly in intimal proliferation to the point of obliteration, hyalinization, and at times necrosis of the blood vessel walls themselves; with many

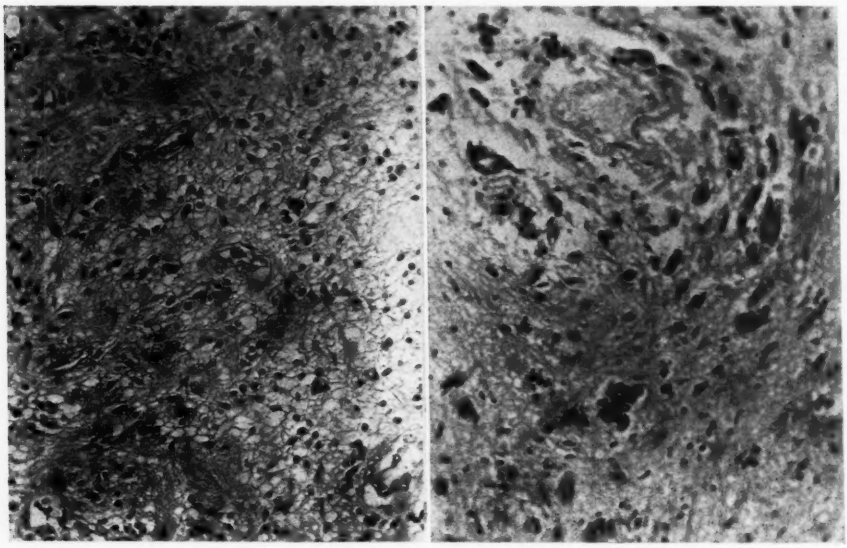

FIG. 5 (*left*). Typical glioblastoma multiforme removed before roentgen irradiation was given. Compare with Fig. 6. (Hematoxylin-eosin, ×230.)

FIG. 6 (*right*). Tumor from same patient as in Fig. 5 after a course of roentgen irradiation. Note changes in parenchyma, stroma and blood vessels. (Hematoxylin-eosin, ×230.)

recent and old thromboses scattered about. However, it is difficult to determine whether these same changes could not occur within the normal range of variation in such a variegated tumor as glioblastoma multiforme at some stage during its lifetime. One tumor without roentgen irradiation, in which microscopic sections were examined first at operation and then at autopsy 7 months later, had changes similar to those which might be construed as evidence of tumor regression, resulting from irradiation therapy.

Davis and Weil[9] also found no real evidence that X-ray had decisively prolonged the survival period of patients beyond the average of those with histologically similar tumors without such therapy. Tarlov[24] noted the fact that histological changes were unimpressive as indications of regression due to irradiation therapy. He, among others, called attention to the stimulative effect of roentgen rays and the possibility of subsequent dedifferentiation

of tumor cells with eventual transition to a more rapidly growing malignant type of glioma, noted particularly at the edges of the tumor mass.

SOCIAL AND ECONOMIC REHABILITATION

Over 1/5 (23) of the patients who survived surgery survived for more than 1 year, and 1/10 (11) for more than 2 years, during which time the patients were not only more comfortable than they had been but were able to fulfill both economic and social obligations. Many of these patients were able to return to work for many months after operation. One patient returned to office work for 9 months after operation, while another returned to work as an executive for 13 months after surgery. The patient who lived for 41 months after surgery became interested in the Oxford Group Movement and was active in this work for most of that time. One patient who had a high-grade papilledema and was blind on entrance to the hospital was operated on and over 100 grams of tumor tissue removed. She made an excellent recovery and her intelligence quotient after operation was 118. She learned Braille and was able to work around the house and care for her two children. She made baskets and leather belts for sale and learned to do dictaphone typing. She divorced her husband, who was a drunkard, and provided for the future care of her children by her parents before she was readmitted to the hospital with a recurrence of her tumor almost 2 years later. She was re-operated upon and almost 100 grams more of tumor tissue removed. She lived for 3 more months.

SUMMARY

Two hundred eleven patients with verified glioblastoma multiforme have been studied. Of these, 187 were operated upon and 110 survived surgery. Twenty-three patients lived for more than 1 year following surgery, and 11 for more than 2 years, during which time many of them were able to fulfill economic and social obligations. On the basis of the microscopic characteristics the tumors were divided into angiothrombotic and angioproliferative groups. The survival time was longer in the latter group. The patients in the extreme age groups had a poor prognosis. The operative mortality was higher if more than one lobe was involved or if the tumor extended into the opposite hemisphere. The patients in whom all or most of the tumor was removed survived for longer periods of time than those in whom only a surgical biopsy was taken. If the bone flap was removed, there were fewer operative deaths than if only the usual decompression was done. There was a longer survival period in those patients who received adequate roentgen irradiation. There were definite microscopic differences before and after roentgen irradiation in 5 tumors. Seven tumors showed a transition from a more benign glioma to glioblastoma multiforme during treatment of the patients.

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ARTERIAL ANOMALIES OF THE SPINAL CORD*

JOSEPH A. EPSTEIN, M.D., AARON J. BELLER, M.D.,
AND IRA COHEN, M.D.

Departments of Neurology and Neurosurgery, The Mount Sinai Hospital, New York, N. Y.

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VASCULAR anomalies of the central nervous system include both arterial and venous malformations. Although found most frequently in the brain, they have also been encountered in the spinal cord and retina. While the venous and arteriovenous abnormalities have received much attention, those of an arterial nature have rarely been described. A review of such lesions in the spinal cord has disclosed 3 reports.[2,4,14] Only in the case of Brasch[4] were satisfactory histopathological studies available. He employed the term, angioma arteriale serpentinicum, to describe his findings. A designation other than "arterial anomaly" as suggested by Wyburn-Mason[14] is unjustified because the inadequate number of postmortem examinations makes it impossible to define the exact nature of the lesion. Names selected because of a gross resemblance to lesions of the brain variously called angioma arteriale racemosum, cirsoid aneurysm, angioma plexiforme and varix arteriale or aneurysmaticus are lacking in specificity and their use only results in error and confusion.

The paucity of reports is surprising in view of the number of these lesions found at operation for spinal cord neoplasm at The Mount Sinai Hospital. Since 1931, 77 intradural, extramedullary cord lesions were exposed. Of this group, 6 could be classified as arterial anomalies. Five cases have been found in the literature after eliminating those of a venous or arteriovenous character. It is felt that similar lesions almost surely have been observed elsewhere but, unfortunately, have never been described. This presentation may act as a stimulus to help establish the arterial anomalies as a nosological entity separating them from the other vascular malformations.

CASE REPORTS

Case 1. A 40-year-old housewife was admitted June 1946 with a 7-year history of sensations of "electricity" in the left leg, occasionally associated with mild low back pain. During the last 4 years intermittent "cramp-like spasms" of increasing severity radiated from the back to the groin. They were aggravated by walking long distances but unaffected by straining. Her pain was worse during menstruation.

Examination. No gross physical abnormalities were disclosed. There was a tendency to drag the right foot. Deep tendon reflexes were increased in the right lower extremity, where Babinski and Chaddock signs were found. Superficial abdominal reflexes were absent on the right. There was a questionable band of hyperalgesia at

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the level of T12 bilaterally and over L2 on the left. Flexion of lumbar spine was limited. There was minimal tenderness on percussion of lumbosacral spine.

Lumbar puncture revealed an initial pressure of 110 mm. of cerebrospinal fluid; manometrics normal; no cells seen; total protein 38 mg. per cent. Roentgenograms showed only slight narrowing of the lumbosacral interspace. Pantopaque myelography disclosed an obstruction at level of upper border of 1st lumbar vertebra. Manometrics repeated the next day again showed no evidence of block. A myelo-

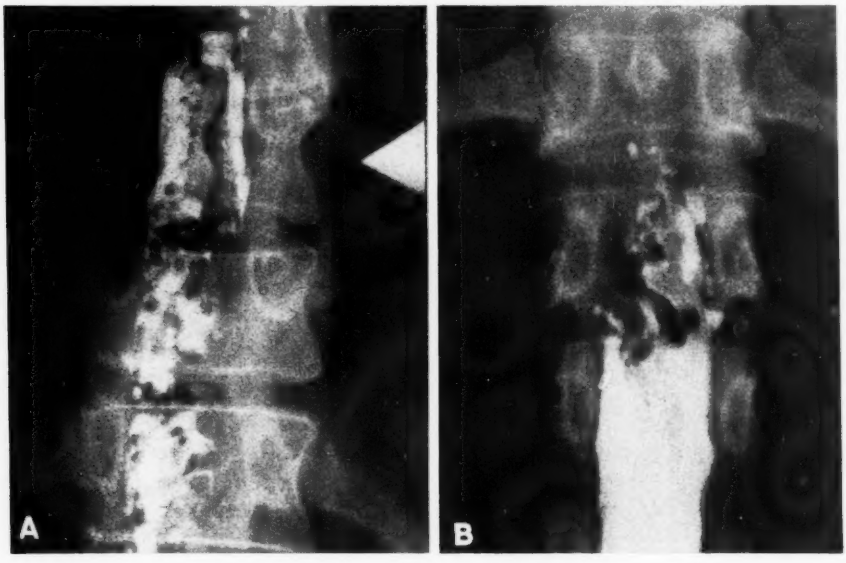

FIG. 1. Myelogram (Case 1) showing curvilinear shadows in the lower dorsal region outlining the anomalous vessels. A. Cisternal myelogram; B. Endolumbar myelogram.

gram, repeated through the cisternal route, showed pantopaque dispersed in a scattered, irregular pattern at the level of lower 3 dorsal vertebrae and 1st lumbar vertebra, suggestive of an arachnoiditis. She was discharged with this diagnosis.

Course. The patient's symptoms persisted and she was readmitted July 1947. Attacks of pain in the low back and left thigh occurred more often and were of greater intensity. There were no sphincteric disturbances. Neurological findings were similar to those noted at previous admission.

Manometric studies indicated a partial block. On being repeated 3 days later, this finding was confirmed. Spinal fluid studies were normal.

Pantopaque myelography revealed an almost complete block at the space between the 1st lumbar and 12th dorsal vertebrae. Here, the oil dispersed and trickled down the left side. Oil injected through the cisternal route was delayed temporarily at the 10th interspace where the column dispersed, forming globular and curvilinear shadows (Fig. 1). While the findings at this time were suggestive of an arachnoiditis, varices or a hemangioma of the cord could not be excluded.

Operation. Laminectomy (Dr. Sidney W. Gross). The spines and laminae of the 10th, 11th, and 12th thoracic vertebrae were removed. The dura appeared normal and was opened, disclosing an extensive vascular malformation on the posterior aspect of the cord extending to the right (Fig. 2). Tortuous coils and loops of vessels, measuring up to 3 mm. in diameter and filled with bright red blood, entwined themselves on the surface and among the nerve roots. The spinal cord seen between the vessels appeared normal. In order to provide an adequate decompression, the dura was left open and a flap of fascia and fatty tissue was sutured over the defect.

Fig. 2. Drawing made from operative sketch of the lesion found in Case 1.

Course. The patient was almost completely relieved of pain. Six days later the ankle and knee jerks were equal and of normal intensity. Although the Babinski sign was equivocal on the right, the Rossolimo sign was markedly positive. There were no sensory changes. It was not considered advisable to give radiotherapy and she was discharged 11 days postoperative. When seen in April, 1948, she was symptom free except for the appearance of pain during menstrual periods and during heavy exertion. A band of hyperalgesia over L1 bilaterally was the only positive finding.

*Case 2.** A 48-year-old Chinese man was admitted October 1947 with a 4-month history of persistent numbness and impaired appreciation of temperature over the entire left lower extremity. Soreness and a "dull pain" between the shoulder blades, of greater intensity on the right, were noted 2 months later. These symptoms were aggravated by activity, and were most severe when the patient was recumbent. About 1½ months before admission, he became severely constipated and could defecate only after enemas. Weakness of the left leg was noted at this time; 2 weeks before admission it spread to the right leg. During the last week, he had great difficulty in voiding, urinating only after prolonged periods and with much stress.

Past history was non-contributory. There were no traumatic incidents.

Examination. There was a soft, non-tender subcutaneous mass, measuring 8 by 6 by 3 cm., in the dorsal region lateral to the spinous process of T5 on the right. It was not fixed to the skin and underlying tissues.

Gait was slightly unsteady, with weakness of right lower extremity. Deep tendon reflexes were increased in both lower extremities, more so on the right. Babinski signs were elicited bilaterally, more actively on the right. Abdominal and cremasteric reflexes were absent. There were no abnormal findings in upper extremities.

* We would like to thank Dr. I. Freiman for permission to present this case.

Sensory examination revealed inconstant, poorly defined levels. There was loss of pain and temperature appreciation from approximately T9 down on the left side including the saddle area.

Spinal fluid pressure was normal; manometrics revealed evidence of a partial block; total protein 50 mg. per cent; no cells.

X-rays of dorsal spine revealed no evidence of intrinsic disease. There was slight hypertrophic spondylitis of lower dorsal vertebrae and scoliosis of upper dorsal spine to the left.

FIG. 3. Myelogram (Case 2) of the cervicodorsal region showing the scattered droplets of pantopaque outlining the vascular malformation in this area. The ease with which this pattern can be confused with that seen in arachnoiditis is evident.

A cisternal myelogram, using 3 cc. of pantopaque, revealed moderate delay in passage of the column of oil at level of 5th cervical interspace. Here it broke up and slowly trickled downwards scattering throughout the lower levels, remaining suspended in particulate and curvilinear fashion in upper dorsal regions (Fig. 3). The findings were suggestive of an arachnoiditis or a vascular anomaly.

The suggestion of a Brown-Séquard syndrome persisted with variation in the upper levels of impaired sensation on the left. During the 9 days that preceded operation, the level varied from T2 to T12, being most persistent in upper dorsal regions. Sphincter disturbances were consistently out of proportion to the slight motor deficiency.

Operation. Laminectomy (Dr. Ira Cohen). Removal of spines and laminae of C6, C7, and T1. The region was not unusually vascular. The dura appeared normal. On opening the dura, numerous coiled and intertwined vessels filled with bright red blood were exposed on the surface of the cord, extending laterally between the nerve roots and varying in size from 1 to 3 mm. (Fig. 4). They appeared to be suspended in the subarachnoid space and did not pulsate. There were no venous dilatations. The upper and lower limits of the arterial anomaly extended beyond the operative exposure. The dura was closed and wound sutured in usual manner.

Course was uneventful and patient was discharged 10 days after operation. Neurological findings had persisted with a sensory level at T3 on the right. There were no reflex changes from those noted at admission. He began to void spontaneously on 7th postoperative day and required no further catheterization.

He was given a course of deep x-ray therapy. After a brief period of relief, his symptoms and signs returned and have subsequently been progressive. When seen

6 months after discharge, there was marked increase in weakness of the lower extremities, a sensory level at T2, and sphincteric disturbances were prominent.

Case 3. A 40-year-old shipping clerk was admitted with a 3-month history of progressive weakness of his lower extremities, at first involving the left and later the right. For the past 2 months, he complained of difficulty in starting urination and of constipation of increasing severity. Coldness and burning sensations were present in both feet. There were no other sensory abnormalities. Past history was not contributory.

Examination. There was marked tenderness on percussion over the spines of 4th and 5th lumbar vertebrae. There was weakness of both legs, the right more than the left. Gait was broad based and the patient fatigued rapidly. Knee jerks were hyperactive, more so on the right, and ankle jerks could not be elicited. Babinski sign was present on the right but equivocal on the left. Superficial abdominal reflexes were absent. Sensory findings were inconsistent and varied considerably on repeated examinations. There was a vague level of hypalgesia and hypesthesia bilaterally from approximately T9-T10 down, most evident over the distal portions of the extremities. Vibratory and position sense were undisturbed.

Lumbar puncture revealed normal pressure and dynamics; fluid contained 5 cells/c.mm.; total protein 59 mg. per cent. Roentgenograms of lumbosacral spine revealed no significant findings. Cisternal myelography disclosed a rapid passage through the dorsal canal. Following endolumbar injection, there was temporary delay in flow at level of lower border of 4th lumbar vertebra. When this was overcome, there was no definite deformity in the canal.

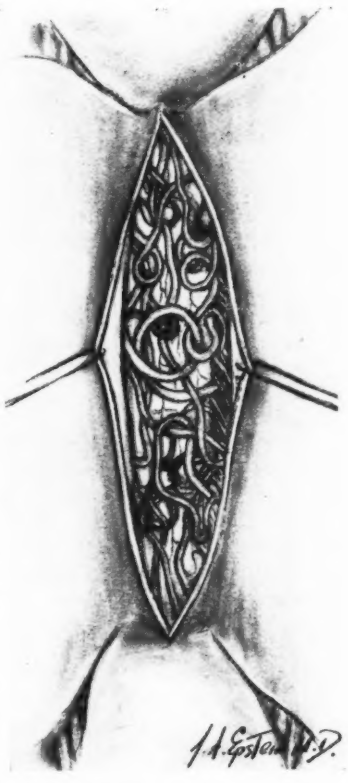

FIG. 4. Drawing made from operative sketch of the lesion found in Case 2.

Within a 2-week period he developed an almost complete paraplegia. Deep tendon reflexes in lower extremities were no longer elicited and plantar responses were inactive. Sensory examination continued to yield varying results. Vibratory and position sense were found to be impaired by some observers, and a poorly demarcated level of analgesia was defined at level of 1st lumbar segment.

Operation. Laminectomy (Dr. Ira Cohen), on 14th hospital day. Removal of spines and laminae of 11th and 12th dorsal vertebrae. On opening the dura tortuous, dilated adventitious vessels, arterial in nature, were exposed overlying the left lateral surface of the spinal cord and extending caudally over conus, epiconus and roots of the cauda equina. A few similar vascular collections were seen on the right

side. A catheter was passed upward and downward for distances of 8–10 cm. without encountering any obstruction. The dura was closed and wound approximated in usual manner.

Course. Operation was followed by a complete flaccid paraplegia, cord bladder and rectal incontinence. Sensory levels were still not definite. There was loss of appreciation of pin-prick up to the 11th thoracic dermatomes. Touch, position sense and pallesthesia were markedly impaired. The patient left the hospital 6 weeks after operation, at which time sacral decubitus ulcers and bladder infection were serious problems. There was no further return of function.

He was transferred to the Montefiore Hospital where he died 7 years later. An autopsy was performed and the histopathological nature of the lesion confirmed.

Case 4. A 54-year-old man was first admitted in November, 1931. During the preceding 8 years, he had two attacks of "lumbago" which subsided spontaneously. Three months before admission he began to complain of numbness, coldness and progressive weakness in both lower extremities, occasionally associated with sensations of "pins and needles." Disagreeable "drawing sensations" involving muscles of the posterior aspect of legs and thighs were also present. Three weeks before admission he noted difficulty in starting the urinary stream and 2 weeks later, involuntary contractions of right lower extremity.

Examination. There was an area of impaired appreciation of heat and cold with hyperpathia below the level of L3 bilaterally. Plantar responses, abdominal and cremasteric reflexes could not be elicited. Deep tendon reflexes were hyperactive in both lower extremities.

Manometrics revealed no block; spinal fluid protein 49 mg. per cent.

Course. The diagnosis of intramedullary cord disease was made and the patient was given non-specific protein therapy with resulting marked recession of subjective and objective sensory changes. The hyperreflexia, however, persisted as did the sphincteric disturbances. He was discharged slightly improved.

Relief was temporary and his symptoms recurred the following month. Occasional episodes of incontinence alternated with the usual picture of urinary retention. Radiotherapy had no beneficial effect. On Mar. 28, 1932 he had an episode of acute retention requiring catheterization. He was unable to completely empty his bladder thereafter. Because of these difficulties, myelography was performed. It revealed partial obstruction at level of 11th thoracic vertebral body.

Operation. Laminectomy (Dr. Jefferson Browder) was performed at another institution on May 5, 1932. The report was as follows: "There was found a very extensive angioblastic malformation with and about the conus extending downward over the upper aspect of the cauda equina. This aneurysmal-like formation of blood vessels was made up for the most part of arteries woven into a snarl-like, pulsating mass. Some of these vessels were as large as the radial artery. They seemed to practically fill the intradural space. Because of the extensive involvement, no attempt was made to coagulate them. The dura was left open."

Course. Following operation, there was fecal and urinary retention. Numbness, paresthesias and inability to walk were associated with involuntary contractions of right lower extremity. After a course of radiotherapy in November 1932, involuntary contractions disappeared and motor power improved. In January 1933, a relapse occurred leaving patient completely unable to walk.

He was readmitted to The Mount Sinai Hospital in March 1933, at which time there was paraparesis with involuntary contractions in right lower extremity, hyperesthesia below 3rd lumbar segment bilaterally and anesthesia in perianal region.

Knee jerks were hyperactive. Ankle jerks could not be elicited. Rectal sphincter was relaxed. Urinary retention with a large residua was persistent. He was discharged in June 1933. There was no improvement during this period. No further information was obtainable.

Case 5. A 62-year-old woman was admitted March 1941 with a 13-month history of dull pain over the lower spine, accompanied by nocturia and urgency of urination. Constipation was a major problem. One week before admission she was awakened by excruciating pain deep in the right buttock which radiated down the thigh. To relieve her pain, she douched herself. In doing this, she discovered that the perineal region was numb and that there was no sensation in the vagina. By the next morning there was involuntary dribbling of urine and inability to stand on her legs, which were numb and weak.

Examination. There was weakness of both lower extremities, more marked in biceps femoris and in dorsiflexors of right foot. Ankle jerks were absent bilaterally. Knee jerks were hyperactive. Neutral plantar responses and active abdominal reflexes were elicited. Vibratory sense was absent up to lower lumbar spine. Position sense was intact. Appreciation of touch, pain and temperature in S3, S4 and S5 dermatomes was absent bilaterally. There was tenderness over lower lumbar spine. Overflow incontinence was present and rectal sphincter was patulous.

Clinical diagnosis: Tumor of cauda equina, probably metastatic.

Manometric study disclosed a partial block; no cells in cerebrospinal fluid; total protein 110 mg. per cent. X-rays of dorsolumbosacral spine showed marked osteoarthritis. Myelography disclosed no abnormalities.

Operation. Laminectomy (Dr. Ira Cohen). Removal of lamina of T12 and L1. When the arachnoid was opened, many abnormal vessels were seen on the dorsal aspect of the lower end of the cord and the conus. Those overlying the conus were bunched to form a small rosette. A diagnosis of arterial anomaly was made, and dura and wound closed.

Postoperative course was smooth but without improvement. Deep radiotherapy was given without any change in her condition.

The patient was seen 10 months later at which time her chief complaints were incontinence of urine and pain in the rectum. She was still alive when last heard from $6\frac{1}{2}$ years after discharge.

Case 6. A 56-year-old man was admitted with a 5-month history of feelings of coldness and heaviness in both feet gradually spreading up to the hips, more marked on the left. During this period he experienced weakness in both legs, the left greater than the right. He also noticed that he did not know the position of his legs and had more difficulty in walking at night than in the day.

Examination disclosed moderate weakness in both lower extremities with slight atrophy of right calf. There were hyperactive knee jerks and absent ankle jerks. Babinski and Chaddock signs were elicited bilaterally. Fibrillations were present in both calves. Absent vibratory and markedly diminished position sense were found in all toes. Hypalgesia was present over L4, L5, S1 and S2 segments. The saddle area was not affected. The clinical impressions included amyotrophic lateral sclerosis, meningovascular lues, combined sclerosis, and intramedullary neoplasm. Manometric study revealed a partial block. Initial pressure was 150 mm.; final pressure after removal of 10 cc. of clear cerebrospinal fluid was 0; 6 cells; total protein 48 mg. per cent.

X-rays of dorsolumbosacral spine showed moderate hypertrophic spondylitis.

Myelogram showed a disturbance in passage of the iodized oil at level of T11 with dispersal of the stream of opaque material.

Operation. Laminectomy (Dr. Ira Cohen). Removal of spines and lamina of T10 and T11. "Immediately on opening the dura there were seen lying on the posterior surface of the cord, opposite T11, two large tufts of abnormal vessels 1 cm. in diameter in addition to many single strands of supernumerary vessels. It was obvious that we were dealing with an angiomatous anomaly. The dura and wound were closed."

Course. Patient was given deep x-ray therapy. When discharged 2½ weeks later there was marked improvement of power in both lower extremities, otherwise neurological status was unchanged.

He was observed in the follow-up clinic for 22 months. During this time there was improvement in motor power; however, sensory abnormalities persisted. He still complained of pain in the back radiating down both legs.

The following 5 cases taken from the literature are abstracted in brief.

The patient of Brasch[4] was a 61-year-old man with a history of progressive weakness of lower extremities for 2 years. He later became incontinent and complained of pain in gluteal region and legs. There was weakness and absence of deep tendon reflexes in lower limbs. Hypalgesia was found over the thighs, and anesthesia more distally. He died 10 days after admission. Postmortem examination disclosed thickened, tortuous arteries coiled over dorsal and ventral surfaces of cord from D5 to L1 and extending into its substance.

The patient (age not recorded) presented by Balck and Harman[2] had an episode of pain, weakness and sensory changes in lower extremities 4 years before admission. Two years later, a similar episode was followed by a brief remission and sudden relapse, this time associated with urinary retention and pain in cervical region as well as in thighs. His temperature rose rapidly and he became unconscious, losing control of his sphincters. Death followed development of nuchal rigidity and small pupils. Postmortem examination showed evidence of subarachnoid hemorrhage extending into cervical region with destruction of a portion of the lumbar spinal cord. An arterial anomaly was present in this region.

The 3 patients of Wyburn-Mason[14] were 55, 39, and 35 years of age. They were men and all showed symptoms of a progressive or intermittent nature with subsequent evidence of transverse lesions in dorsal region of spinal cord. Symptoms had been present for 8 months, 2 years and 3 years respectively. Manometric study performed in 2 cases disclosed evidence of a partial block in 1; spinal fluid protein was elevated in 2. Roentgenograms of spine were essentially normal. Myelography showed a block at D6 in the 1st case and no obstruction in the 2nd. The lesion was demonstrated at operation in each case, in the 1st and 2nd at middorsal levels and in upper dorsal region in the 3rd.

Of the entire group, 9 were men and 2 were women. The youngest was 35, the oldest 62 and the average age was 54. The occurrence of arterial anomalies about 4 times more frequently in males than in females is similar to that seen in the venous abnormalities and is unlike that of the arteriovenous type in which the incidence is almost equal.[14]

The symptoms were progressive in nature without remission in 7 cases and were intermittent with gradually developing signs of progression in 4.

In the first group, the duration of the illness was comparatively short, varying from periods of 1 to 8 months to a maximum of 2 years. In the second group, the duration was from 3 to 8 years. In 1 case, progressive symptoms of 1 month's duration were climaxed by a sudden hemorrhage into the subarachnoid space with signs of an acute transverse myelopathy.

Noteworthy was the absence of cutaneous naevi. This is in contrast with the numerous reports of skin hemangiomas in the dermatome distribution of segments of the spinal cord involved by a venous or arteriovenous anomaly. Congenital malformations were nowhere in evidence. A subcutaneous lipoma was found in 1 case. In general, the physical findings have not been significant. In no case was the blood pressure abnormal. Percussion tenderness of the spine overlying the lesion was noted in 2 cases. The neurological examination was characterized by sensory levels that shifted during the same and on subsequent days. Findings of progressive cord damage with final evidence of a complete transverse myelopathy were the presenting features or followed in the wake of recurrent attacks. There was a suggestion of a Brown-Séquard syndrome accompanying the lesion of the cervical cord. Five patients showed signs of a tumor of the cauda equina. The others manifested evidence of intramedullary and extramedullary cord disease at the dorsal levels. Varying signs of root irritation were present. These findings resulted in numerous diagnoses such as tabes, amyotrophic lateral sclerosis, combined sclerosis and, most frequently, neoplasm.

The serology was negative in every case of the present series. Study of the cerebrospinal fluid revealed slight to moderate elevation in the total protein. In the 6 patients of our series, the protein was above normal, reaching a maximum of 110 mg. per cent. Manometric tests disclosed a partial block in 5 cases with no evidence of obstruction in 2. In 4 cases, no information was available. Myelography showed no evidence of block in 2 cases, a partial block in 5 and a complete block in 1. In the 2 cases of our series in which pantopaque was used through both the cisternal and endolumbar routes, fluoroscopy was performed by Dr. W. Newman. It disclosed an interruption in the column of oil at the upper level of the lesions with a subsequent dispersal into small rivulets and droplets whose outlines formed a pattern defining the course of tortuous, convoluted vessels, at the same time vaguely suggesting arachnoid adhesions. Roentgenograms showed with clarity the defects suggested during fluoroscopy. The findings were considered to be pathognomonic of a vascular malformation. This is the only means by which a preoperative diagnosis may be ventured with assurance.

Laminectomy was performed in 9 of the 11 cases. The subcutaneous tissues and muscles were not unusually vascular. The epidural fat was present in normal amounts and no extradural abnormalities were encountered. The dura was of normal color and under no unusual tension. Nothing abnormal could be discerned through it. On opening the dura, the lesion was exposed. It consisted of convoluted, intertwined vessels filled with bright red blood and measuring up to 3 mm. in diameter. The vessels coiled around and be-

tween the nerve roots extending laterally and ventrally, at times filling the entire space between cord and dura. Pulsations were seen in 1 case. Neither the upper nor the lower limits were exposed. The spinal cord seen between the vessels appeared to be of normal size and color and no evidence of cord compression or swelling was observed. No vessels filled with venous blood contributed to the malformation.

The dura was left open in 3 cases and was closed in 4, no record being available in the remaining cases. Two in the present group showed improvement, 1 followed for a period of 2 years after operation, the 2nd for 10 months. In the former, the dura was closed and radiotherapy was instituted. In the latter, the dura was left open and a fat-fascia pellicle graft inserted into the defect. No radiotherapy was given. This patient was relieved postoperatively of the excruciating pain which formerly was her chief complaint.

The lesion occurred with equal frequency in the lumbosacral and dorsal segments of the cord, 1 being found in the cervicodorsal region.

Unfortunately, postmortem examination was performed in but 3 of the entire series. An adequate histopathological study is available only in the case reported by Brasch[4] in which the arterial malformation extended from T5 to L1. A greatly enlarged vessel from which the malformation had its origin pierced the dura at T5 and formed numerous coils and loops on the dorsal surface of the cord. On cross section, the enlarged surface vessels showed a concentric medial hypertrophy with preservation of a single intimal layer and a normal adventitia. The tunica elastica was interrupted and fragmented. Numerous small arteries penetrated the cord at its dorsal surface in the region of the posterior roots. These also showed the same medial hypertrophy with occasional obliteration of the lumen of the vessel by concentric thickening of the tunica muscularis. A well preserved internal elastic membrane normally absent in arteries of this size was prominent.

Multiple small extravasations were present in the lumbar segments of the cord. Specific stains disclosed degenerative changes in the cord substance and included both the grey and white matter, mainly involving the dorsal columns.

In the case described by Balck[2] evidence of fresh hemorrhage was discovered on opening the dura. The right half of the cord in the lumbar region was destroyed. On the dorsal surface of the cord, an "angiomatous malformation" was found which extended laterally. The vessels were large and thick-walled. Rupture had occurred with a resultant massive hemorrhage into the subarachnoid space extending up to the cervical region. There was no evidence of malignancy. In the only case of the present series in which autopsy was performed, 7 years after the patient was transferred to the Montefiore Hospital, the arterial nature of the lesion was confirmed.

On a symptomatic basis and by examination, it is impossible to differentiate the arterial anomalies from the venous and arteriovenous malformations. The same fluctuation in symptoms, the varying sensory levels and the evidence of intramedullary and extramedullary cord disease with root

compression may exist in all. The absence of cutaneous vascular naevi in the present group can be of little assistance in the differentiation of the arterial malformations from the others, although its occurrence in association with the venous, the arteriovenous lesions and hemangiomas is quite frequent.[5,6,7,9,10,14] The presence of a lipoma is also of no aid in differentiation as it may occur in all three vascular anomalies.[3,7,9]

The evidence of a partial block on a manometric study, the pathognomonic findings on pantopaque myelography and the normal roentgenogram of the spine are shared by all. In conjunction with each other, these findings serve to differentiate this group from cord neoplasm, disseminated sclerosis, inflammatory lesions of the cord, thrombosis of the anterior spinal artery and syringomyelia. The narrow curvilinear shadows in the myelogram distinguish these vascular lesions from spinal arachnoiditis in which droplets and ragged linear collections are suspended in the subarachnoid space.

It is of significance that certain veins of the spinal cord may attain huge varicose proportions and produce symptoms similar to those of the other arterial and venous malformations. They may be distinguished by their unusual size and resultant displacement of iodized oil over a broad serpentine course.[8]

DISCUSSION

The malformations of the spinal cord have been compared to similar lesions found on the surface of the brain. Although a distinctive group has been classified as predominantly arterial, rarely has the histological structure been adequately confirmed. Cushing and Bailey[6] described a vascular malformation which they preferred to call angioma arteriale. Although the vessels possessed a thickened intima, and a definite internal elastic lamina, they were thin-walled and lacked the muscular layer so prominent in the lesions of the spinal cord. Therefore, it is impossible to consider them to be identical. Because both arteries and veins entered into the primary composition of the lesion in the brain, the name angioma arteriale et venosum as suggested by the same authors would be more exact.

An arterial angioma described by Asenjo and Uiberall[1] at the base of the brain resembles more closely the anomalous vascular formation of the cord. The presence of arterial and at times of undifferentiated vessels forming both efferent and afferent trunks justifies the name arterial angioma. However, medial thickening, which is the outstanding feature of the cord lesion, is lacking, thereby making it difficult to establish identity. After reviewing the so-called arterial angiomas of the brain, in no case has a malformation with all of the characteristics of the lesion in the spinal cord been found.

In 1 case of an aneurysm of a retinal artery described by Theodore and Bonser,[13] the optic disc was obscured by a convoluted, serpentine dilatation of the inferior temporal vessel which was definitely arterial in structure and did not pulsate. Nothing further could be determined as the lesion was asymptomatic.

Evidence of hemorrhage is not unusual in cases of arteriovenous anomalies

of the brain and spinal cord.[3,6,11,12,14] Such hemorrhages undoubtedly contribute to the clinical picture, especially where evidence of massive injury has occurred. The constriction of small vessels by the concentric thickening of the media with eventual occlusion is the probable cause of the fluctuating symptomatology and the persistence of signs of cord injury. It is difficult to evaluate the variation in the signs of root compression following changes in position. A change in the vascular dynamics on standing with probable relief of tension on the nerve roots entwined with the vessels may explain this phenomenon.

Of surgical importance in the exposure of an arterial malformation is the relatively normal state of the subcutaneous tissues and bone overlying the lesion. The dura is neither distended nor discolored. In the venous and arteriovenous malformations, the overlying structures are often excessively vascular and the dura unduly tense transmitting the bluish discoloration of the underlying veins.

Radiotherapy has little effect on the course. Because of the possibility of further occlusion of the small nutrient vessels, it is not considered advisable. Any operative procedure other than decompression would be disastrous in view of the character of the lesion.

SUMMARY

Six cases of arterial anomalies of the spinal cord have been presented with a review of the 5 cases already reported. The distinguishing clinical, roentgenographic and histopathological characteristics are discussed and evaluated, due consideration being given to similar vascular malformations in the brain and retina.

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THE END-RESULTS OF SURGERY FOR RUPTURED LUMBAR INTERVERTEBRAL DISCS

A FOLLOW-UP STUDY OF 327 CASES

R. GLEN SPURLING, M.D., AND EVERETT G. GRANTHAM, M.D.
Department of Neurosurgery, University of Louisville School of Medicine, Louisville, Kentucky

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BETWEEN January 1, 1939, and March 31, 1947, 378 patients were submitted to operation for ruptured lumbar intervertebral discs in the private practice of the authors of this communication. Of these, 260 were operated on between January 1939 and April 1942 and the remaining 118 between January 1946 and March 1947. The four-year break in the series was occasioned by the absence of both authors in military service.

One of the 378 patients died. He was operated on in January 1939 and died on the 10th postoperative day. The clinical cause of death, bilateral bronchopneumonia, was verified by necropsy.

The purpose of this communication is to report the end-results to the present time of 327 of the 377 patients who survived operation.

MATERIALS AND METHODS

In July 1947 a questionnaire concerning the results of their operations for ruptured lumbar intervertebral disc was sent to 363 of the surviving 377 patients operated on during the approximately eight-year period ending in March 1947. The 327 replies that were received represent an approximately 90 per cent response. The 80 patients who took advantage of the opportunity for a follow-up examination offered in the questionnaire constituted, in effect, a control group in that they served as a check on the reliability of the answers given by the whole group.

The questionnaire sent to these patients (Fig. 1) was worked over until the questions had been made so simple that they could not possibly be misunderstood, while at the same time they were designed to supply practical, precise information as to the success of the operation in the individual case.

The break in the series of operations occasioned by the military service of the writers naturally divided the group into long-term and short-term follow ups. When the replies to the questionnaires were tabulated, however, it was found that the replies to the four questions in the long-term group of cases differed from those in the short-term group by less than 1 per cent. In the analysis of results the two series are therefore combined.

It should be emphasized that this follow-up study concerns only private patients. Clinic patients are notoriously difficult to trace and for this reason, if no other, it seemed wise to exclude the entire category.

PRINCIPLES OF MANAGEMENT

For a better comprehension of the results to date in these cases it is necessary to summarize briefly the broad policies that are employed in the man-

agement of patients with suspected ruptured intervertebral discs in the lower lumbar region and which have been set forth in detail in a recent publication.[2] These principles of management have been employed with scarcely any modification in the period covered by this study. In particular, the criteria by which patients are selected for operation, as well as the technical surgical procedure employed, have scarcely varied during the entire period.

The principles of management may be summarized as follows:

1. Unilateral (occasionally bilateral) sciatica complicating low back disability and refractory to conservative treatment is regarded as the only specific

We are attempting to evaluate the results of our surgical efforts and would appreciate a careful reply to this questionnaire.

(1) *Leg Pain:*

a. Relieved completely _____

b. Occasional recurrence (describe) _____

c. No improvement _____

(2) *Back Pain:*

a. Relieved completely _____

b. Occasional recurrence (describe) _____

c. No improvement _____

(3) Are you able to do your usual work? Please give the nature of your present work and any other physical activities in which you participate.

_____

_____

_____

(4) Do you consider your operation to have been successful? (Explain)

_____

_____

_____

If you would care to make an appointment at our office, 405 Heyburn Building, for a check-up we will be glad to make this examination without charge. *A reply within one week will be very much appreciated.*

Name _____

Address _____

Date _____

FIG. 1. Questionnaire sent out to patients operated on for ruptured lumbar intervertebral discs.

indication for surgery on the diagnosis of ruptured lumbar intervertebral disc. Low back pain not complicated by sciatic radiation is practically never accepted as an indication for surgery.

2. The initial episode of pain is treated by conservative measures over a long period of time, since a large proportion of patients with ruptured lumbar intervertebral discs will recover without operation. Surgery is never performed until conservative treatment has been given a fair trial.

3. Most patients with ruptured lumbar intervertebral discs present unequivocal neurologic signs if they have symptoms severe enough to justify operation. These signs are essential for the making of an accurate diagnosis and can also be relied upon for localization of the lesion.

4. The diagnosis of ruptured lumbar intervertebral disc is made on clinical findings. If symptoms are incapacitating, and if clinical findings are not

sufficiently clearcut to localize the lesion accurately, myelography is employed. This is a valuable localizing measure but it is not a diagnostic procedure and is not so regarded.

5. Operation is never performed during a remission, even if the patient presents an absolutely classic history of ruptured lumbar intervertebral disc. Failure to observe this precaution is one of the most common causes of negative explorations, of which more will be said later.

6. Operation for removal of a ruptured lumbar intervertebral disc is accomplished without destruction of the lamina or of the articular facets. Complete removal of the entire nucleus pulposus, loose pieces of annulus fibrosis and cartilaginous plates is possible through a small unilateral interlaminar exposure.

7. The posterior annulus fibrosis is not opened unless a complete or incomplete tear can be demonstrated in it by inspection or by palpation with a blunt instrument. If a tear cannot be demonstrated, the exploration is listed as negative. Adequate decompression of the involved nerve root is, however, carried out.

8. Primary spinal fusion is never combined with simple removal of the ruptured intervertebral disc. Patients who continue to have incapacitating backache after simple disc surgery may require secondary spinal fusion, but a properly performed operation on the disc does not in any way interfere with subsequent fusion operations.

9. Convalescence is strictly supervised. The patient remains recumbent for 10 days after operation, is permitted to walk on the 13th day, and may leave the hospital on the 14th day. Unless there is persistent disability referable to the lower back, no support for this region is regarded as indicated during convalescence. Patients who do sedentary work may resume their activities 4 weeks after operation but laborers should not return to their regular work for 3 to 6 months.

GENERAL DATA

The age range in the 378 patients in this series was from 16 to 74 years. Four patients were under 20 years of age and 18 were over 60 years. There were 259 patients, more than two-thirds, who were between 30 and 50 years of age. The series included 257 men and 121 women.

It is unfortunate that data are available as to the occupation of the patients in only 286 cases. Eighty-six (30 per cent) in this group were laborers, 41 (14 per cent) were farmers, 67 (20 per cent) were housewives, and the remaining 92 patients (36 per cent) were variously students, teachers, office workers, executives, nurses, physicians and salesmen. If farmers are classified as doing heavy work, 44 per cent of the entire group of 286 patients whose occupations are known fell into this group. The housewives, who comprised 20 per cent of the patients, constituted another group whose work requires a great deal of activity even if it cannot properly be termed heavy.

In 5 of the 378 cases there were two ruptured discs, the ruptures in each instance occurring in the 4th and 5th lumbar discs.

Negative Explorations. In the long-term group of cases, in which operation was carried out before 1942, there were 28 negative explorations. In the short-term group, in which operation was carried out after 1945, there were only 2 negative explorations. The difference is partly explained by the fact that in the short-term group not a single patient was operated on during a remission of sciatic pain.

Another significant observation in this connection is that 16 of the 30 patients whose explorations were negative (53 per cent) were submitted to operation after diagnosis had been made and localization accomplished chiefly by myelographic findings rather than by clearcut clinical findings. Moreover, in the remaining 14 cases clinical and myelographic findings were often not in agreement. Apparently the best way to reduce the number of negative explorations is to avoid compromise with the classical clinical symptoms of ruptured lumbar intervertebral disc.

FOLLOW-UP RESULTS

Relief of Pain. Of the 327 patients who replied to the questionnaire 152 (46.5 per cent) stated that they no longer suffered from pain in the leg. Twenty-seven had continued to have pain as severely as before operation. Of the remaining 148 patients (45 per cent), 123 had infrequent attacks of pain which was usually fleeting and was not disabling. The other 25 always had some discomfort and also had attacks of severe pain at varying intervals.

There were 131 patients (40 per cent) who stated that they were completely relieved of the low back pain from which they had suffered before operation. Twenty-five (7.7 per cent) complained that it was just as severe as before operation. The other 171 patients (52.3 per cent) suffered from attacks of low back pain at varying intervals. In some instances recurrences were regular and disabling. In other instances the episodes consisted only of a transient "catch" in the back, usually following heavy lifting or some out-of-the-ordinary twisting motion.

Ability to Work. Of the 327 patients who replied to the questionnaire, 280 (85.6 per cent) stated that they were then doing precisely the same type of work as they had been engaged in prior to the onset of illness. Thirty-nine patients (12 per cent) were still gainfully employed but had had to change to a less strenuous type of occupation, and only 8 patients (2.4 per cent) replied that they were unable to do any type of productive work because of the persistence of symptoms.

Success of the Operation. Two hundred fifty-nine patients (79 per cent) stated that from their standpoint the operation was entirely successful and 36 (11 per cent) regarded it as partly successful. In other words, 90 per cent of the 327 patients were partially or entirely satisfied with the results of surgery.

Composite Results. When the replies to the various inquiries in the questionnaire were tabulated and evaluated as a whole, it was found that 131

patients (40 per cent of those who replied) considered themselves cured. Their answers indicated that they had been relieved of all symptoms, that they had returned to their former occupations, and that they were able to indulge in all the activities, including the recreational activities, to which they had previously been accustomed. Another 128 patients (39.2 per cent) had some residual low back pain or sciatic disability or both but were able to engage in their usual occupations and activities and considered the operation a success. From their personal standpoint, therefore, the results of surgery were apparently satisfactory. In the remaining 68 patients (20.8 per cent) results varied from partial success to complete failure.

Control Group. Each of the 80 patients who returned for follow-up examination in response to the invitation on the questionnaire, and who were regarded as forming a sort of control group, was personally seen and examined by one or the other of the writers. In each instance a detailed follow-up history was obtained, and a careful physical and neurologic examination was made. Twenty-one of the 32 patients who had indicated in their replies that they regarded their operations as unsuccessful were among those who returned to the office. In only three of the 21 was it possible to determine from physical and neurologic signs that the operation had been really unsuccessful, and in none of the patients who returned was the condition found to be worse than the replies to the questionnaire had indicated.

Compensation Group. Of the 378 patients operated on for ruptured lumbar intervertebral disc, 91 (24 per cent) were eligible for compensation for their condition through the Workmen's Compensation Act or through liability insurance companies. Seventy-five of the 91 (82.4 per cent) returned their questionnaires: 48 (64 per cent) stated that they were engaged in the same type of work as before operation; 19 (25.3 per cent) had changed their occupations because of persistent symptoms; and 8 (10.7 per cent) stated that the severity of their symptoms made it impossible for them to work at all. Fifty of the 75 (66.6 per cent) considered the operation successful, 8 (10.7 per cent) considered it partially successful, and 17 (22.7 per cent) considered it as a failure.

Recurrences. Twenty-one (5.5 per cent) of the 378 patients in this series were subjected to secondary operations for suspected recurrence of the ruptured lumbar intervertebral disc. In 10 (2.6 per cent of the total number) true recurrences were demonstrated at operation at the same interspace at which the original lesion had occurred. In 2 other patients ruptured discs were demonstrated at different levels. One of these patients had been operated on in 1942 and had had relief of symptoms until 1946. The second had been operated on in 1939 and had had relief of symptoms until 1946.

In 9 instances re-exploration demonstrated neither a recurrent nor a new lesion, the only finding being dense scarring about the involved nerve root. In no instance was a third recurrence demonstrated or suspected.

Negative Explorations. Of the 30 patients in whom operation had produced only negative findings 18 replied to the questionnaire, and 13 of these (72 per cent) stated that they had been free of symptoms since operation.

DISCUSSION

From the standpoint of the desired objective, information as to the present status of patients operated on for ruptured intervertebral disc, the simple questionnaire used in this study was entirely satisfactory. Some criticism, however, has been raised concerning the question as to whether the operation was considered satisfactory, on the ground that truthful answers cannot be secured to such an inquiry. It is quite true that some patients might be inclined to classify an operation as successful because they survived it. It is also true that others might be so grateful for any degree of relief from disabling pain that they used the term success rather loosely. On the other hand, these were private patients, who had elected operation for relief and who for the most part had financed their own treatment. It seems fair, therefore, to permit them to be the final judges of what treatment achieved, since they alone could evaluate the pain and disability that they had experienced before operation in terms of the postoperative residual or lack of residual.

Probably the most important inquiry in the questionnaire concerned the ability of the patient to do the same work after operation as before his illness. Certainly this is of paramount concern to the individual and it is of equal concern to employers, who might question whether it is sound economic policy to authorize surgery upon employees with disabilities caused by ruptured lumbar intervertebral discs. It is significant, therefore, that more than 85 per cent of the patients who replied to the questionnaire stated that they could do the same work in which they had previously been engaged, and that only 8 patients replied that they could do no work at all because of persistent symptoms. The large proportion of patients engaged in heavy work makes these figures even more significant.

As has been noted, the incidence of dissatisfied patients in the compensation group was twice as large as in the whole group. A detailed discussion of the discrepancy would involve factors beyond the scope of this paper, but the implications of the poorer results in the compensation group are obvious. The same disproportion was evident when these patients were asked to evaluate the success or failure of the operation: 17 of the 32 patients who thought the operation a failure were patients in the compensation group. Campbell and Whitfield[1] had a similar experience. In their 122 followed-up cases of ruptured intervertebral discs, patients receiving compensation accounted for 19 of the 21 failures. Prince was perhaps not unduly cynical when, in the discussion of this communication, he accounted for the disproportion on the basis of the "universally deteriorating effect of insurance on the ethics of the human race." Be this as it may, it is an agreeable surprise to observe the regularity with which patients submitted to surgery for ruptured lumbar intervertebral discs are able to return to their former occupations.

The age of the patient appeared to influence the results of surgery in only two ways: Convalescence was moderately prolonged in patients over 50

years of age, and those in this age group had more residual backache than younger subjects, probably because arthritic changes had developed at the site of the diseased discs. There was no notable difference in results from the standpoint of sex.

It is interesting that 13 of the 18 patients with negative findings at operation who replied to the questionnaire reported relief of symptoms. The explanation is probably to be found in certain points of technique. The first is that in the writers' practice the annulus fibrosus is never opened unless a distinct tear can be demonstrated by inspection or by palpation; even if a disc appears prominent or unusually soft, the annulus is not incised unless a tear, which is the essential feature of the disc lesion, can be demonstrated. The second point is that in all negative explorations the nerve root is thoroughly decompressed laterally into the foramen. It is quite possible that some of the patients who reported relief after operation had a true rupture of the annulus fibrosus which was, however, located so far posterolaterally that it could not be visualized. Under these circumstances a thorough decompression of the nerve root might well account for the amelioration of symptoms.

That the operative treatment of ruptured lumbar intervertebral discs resulted in only 40 per cent of completely cured patients in a series of 327 followed-up cases at first glance might seem discouraging. Upon reflection, however, there seems no reason for pessimism. It may be stated unequivocally that once a lumbar intervertebral disc has ruptured, no surgical effort can hope to restore its anatomic integrity. If this premise be accepted, then the relatively low percentage of complete clinical cures is easy to understand.

Each one of the intervertebral discs in the lower lumbar region is an important weight-bearing structure, so important, in fact, that it is in some respects remarkable that patients learn to compensate for the permanently diseased joint as well as they do. If to the potential disability which is present as the result of the diseased disc is added the additional deleterious factor of nerve root compression from either disc fragments or postoperative infiltration of the nerve roots with fibrous tissue, the clinical results become still more satisfactory. Moreover, as has been pointed out, the functional results are even more gratifying than the number of complete cures secured.

Perhaps the chief value of this follow-up study is the demonstration of the results that can be secured by treating all patients by a single method, simple removal of the injured disc, without any type of spinal fusion. The combined operation, with its inevitable distribution of responsibility for the end-results, does not seem to be in the best interests of the patient at the time the disc operation is performed. If it proves necessary, it can be done as a secondary, independent procedure. A skilfully performed disc operation does not in any way interfere with the operative field if it is decided that persistent symptoms warrant spinal fusion.

The results in this series seem to substantiate this point of view. Assum-

ing that spinal fusion by any method could secure results which were 100 per cent effective—such figures, of course, are beyond the claims of even the most enthusiastic advocates of the procedure—4 out of every 10 patients in this series who recovered completely as the result of a simple disc operation would have been submitted to a needless operation, with its attendant risks to life and with its prolonged period of postoperative disability. Unless a large series of long-term results can be published showing a higher percentage of cures and more satisfactory results with the fusion operation than were secured in this series by simple disc surgery, it seems fair to conclude that the best method of treatment of ruptured lumbar intervertebral discs refractory to conservative measures is by simple removal of the damaged disc.

So far as could be determined from the analysis of these replies, patients regard recurrent low back pain after operation as more troublesome than recurrent sciatica. Patients who regard operation as not satisfactory because they must protect their backs when they are engaged in any type of heavy work or exercise form the group in which secondary fusion operations might perhaps be recommended. It is of interest, therefore, that no patient in this series who suffered from recurrent back pain considered his disability sufficiently severe to warrant fusion for its relief.

SUMMARY AND CONCLUSIONS

1. A follow-up study of 378 patients submitted to operation for ruptured lumbar intervertebral disc achieved a response from 327 patients, 80 of whom were also examined personally. Operation in each case was limited to simple removal of the diseased disc.

2. Forty per cent of the patients who replied to the questionnaire could be regarded, on the basis of their own testimony, as completely cured, while another 39 per cent had more or less satisfactory results. In the remaining 21 per cent the results were only fairly successful or operation had to be regarded as a failure. More than 85 per cent of the patients, however, were able to return to their previous occupations, while only 2.2 per cent were unable to work at all. The disproportionately large number of less than satisfactory results in the patients receiving compensation was noteworthy.

3. This series of cases shows the high proportion of cures and of satisfactory functional results that can be secured with a simple operation on the ruptured disc. Spinal fusion is therefore regarded as contraindicated at the original operation, and it is recommended that it be reserved as a secondary procedure for those cases in which persistence of symptoms warrants additional surgery.

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SELECTIVE CORTICAL UNDERCUTTING AS A MEANS OF MODIFYING AND STUDYING FRONTAL LOBE FUNCTION IN MAN

PRELIMINARY REPORT OF FORTY-THREE OPERATIVE CASES*

WILLIAM BEECHER SCOVILLE, M.D.†
Hartford, Connecticut

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THE physiological function of the frontal lobe in man still remains an uncharted field. A technique of cortical undercutting has been devised to enable one to detach certain areas of the frontal cortex by making a ne of cleavage at the relatively avascular junction of gray and white matter y means of a suction tip and spatula, thus interrupting the long association bers of the overlying cortex. The short U association fibers to adjacent gyri re interrupted by angling the undercutting superficially into the gray natter at the peripheral borders of the area to be isolated. Cortical undercutting thus constitutes a method of producing discrete cortical lesions vithout interference of the blood supply to adjacent areas of the brain. Forty-three operative cases have established the technical feasibility of this nethod. In addition to preservation of the blood supply, undercutting has other surgical advantages over cortical ablations. It can be accomplished under direct vision through short straight cortical incisions without leaving a raw exposed surface. Interruption of bridging veins and large arterial branches is unnecessary. Ligatures and electrocoagulation are used only in the initial straight incision. Undercutting can be carried out in areas where ablation is impossible. Reasonably exact delineation of the undercut area can be made with steel or tantalum wire loops.

SELECTION

Three areas have been selected for cortical undercutting: partial Brodmann's areas 9 and 10, the orbital surface and the cingulate gyrus. Areas 9 and 10 have been selected because of a favorable alteration of human behavior demonstrated by Penfield,[6] Pool[2] and Freeman.[3] The orbital and cingulate gyrus areas have been chosen because of their obvious importance in animal experimental work.[1,4,5,7,8] Undercutting of areas 9 and 10 is roughly equal in extent to the areas ablated by Penfield and Pool in their gyrectomy and topectomy operations respectively. Undercutting of the orbital surface includes the entire orbital surface, especially the prechiasmal area 47. Undercutting of the cingulate gyrus includes the entire anterior limbic area together with Brodmann's area 32 and small medial portions of areas 9 and 10.

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* In conjunction with the Connecticut Cooperative Lobotomy Study.

† 56 Garden Street, Hartford 5, Connecticut.

CASES

Forty patients have undergone cortical undercutting for the treatment o mental disease at the Connecticut State Hospital, Institute of Living, an Norwich State Hospital, including 19 areas 9 and 10, 11 orbital, and 1 cingulate gyrus undercuttings. Three patients have undergone undercuttin of areas 9 and 10 for intractable pain at the Hartford Hospital. These patients have been followed for from 1 to 8 months.

TECHNIQUE

Certain instruments are necessary for the cortical undercutting operation (Fig. 1). 1) A dental headrest for application to a standard table or stretcher.* 2) A

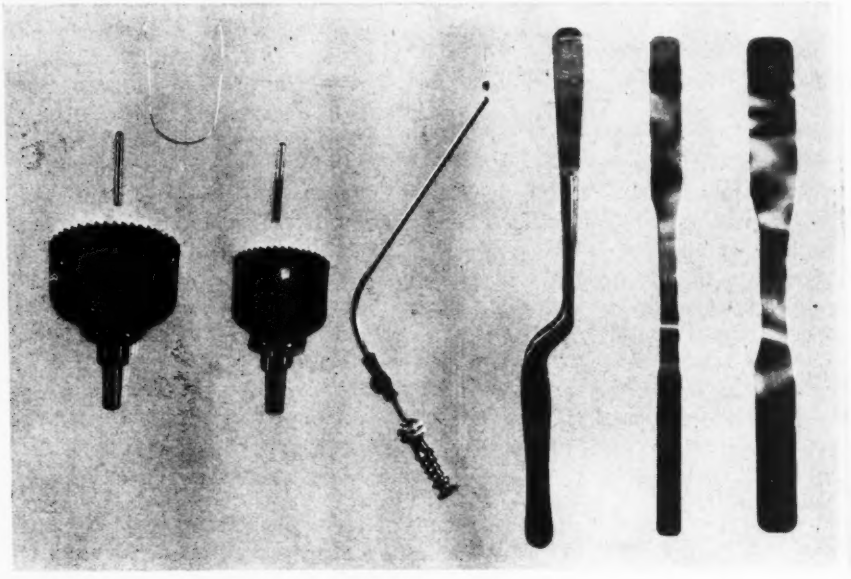

FIG. 1. Instruments used in cortical undercutting.

headlight is essential. 3) Self-retaining scalp retractors. 4) $1\frac{1}{2}''$ and $2''$ trephines can be modified from machinists' standard "hole saws" which can be purchased fo $1.00 from machine tool or auto supply stores. They are fitted with a centering point and shank for use with a standard Hudson drill handle.† A separate $\frac{1}{8}''$ twist drill is used for making the centering hole for the trephine points. 5) A fine No. 1 suction tip, to which is attached the electrocautery wire.‡ It is necessary that this be the

* Approximate cost $17.50 by General Electric Company.

† Complete assembly by local machinist c/o the author can be purchased for $10.00 for the trephine and $15.00 for spatula-forceps.

‡ Manufactured by Codman & Shurtleff, Surgical Supply House, Boston, Massachusetts.

smallest caliber, measuring 2–3 mm. in total diameter. 6) Narrow and medium width, flat, flexible brain spatulae.‡ 7) Loops of 30-gauge stainless steel or tantalum wire are used for delineation of the area of undercutting. 8) Large size bayonet forceps, the prongs of which have been replaced by flat, 1.5 cm. wide brain spatulae.† When inserted into a cortical incision and allowed to expand, these spatula-forceps give an excellent exposure for the undercutting of the medial and orbital surfaces of the brain.

X-rays are taken to show the coronal suture and frontal sinuses. Penicillin and sulfadiazine are used prophylactically. Seventy-five per cent of the cases have been done under local novocaine infiltration plus 4.5 gr. nembutal and 75 mg. demerol in order to permit the carrying out of physiological experiments. Pentothal is used on occasion without intubation and the chin is held up by adhesive tape attached to the overlying table.

I. CORTICAL UNDERCUTTING OF PARTIAL BRODMANN'S AREAS 9 AND 10

Patient lies supine at shoulder height with head flexed sharply forward. A 6″ straight scalp incision is made parallel and $1\frac{1}{2}$″ anterior to the coronal suture, and the skull is exposed subperiosteally over a considerable area in front of the coronal suture. Bilateral 5 cm. buttons of bone are removed with large-sized trephines with their medial edges situated 5 mm. on either side of the midline and their posterior edges at a level with the midline coronal suture. Straight dural incisions are made at right angles to the falx 2 cm. anterior to the midline coronal suture. A small L extension of the dural incision is carried forward along the lateral borders of the bone buttons (Fig. 2). Bilateral straight 3 cm. cortical incisions are made under the dural incisions at right angles to the falx. An additional check on the location of the posterior edge of the undercutting can be made by inserting a ventricular needle stylet into this incision and sliding it forward under the cortex until it impinges on the forward boundary of the floor of the anterior fossa. This distance should measure approximately 7 to 7.5 cm. (Fig. 3). Using the headlight, flat brain spatulae and *fine* suction tip, the demarcation between gray and white matter is identified at a depth of 1 cm. and the gray matter is undercut for a distance of 6 to 7 cm. anteriorly (to the tips of the frontal lobes) and 3.5 to 4 cm. laterally (Fig. 4). The deep superior frontal sulcus, becoming more shallow anteriorly, is encountered and must be undercut in order to obtain the full width of 4 cm. After completion of the undercutting of the superior convexity, the undercut cortex is elevated by a wide spatula held by an assistant and a second narrow spatula is used to make a right angle corner along the medial surface of the frontal lobes to a depth of 2 cm. This is accomplished with the narrow spatula and fine suction tube and carried down approximately to the cingulate sulcus (Figs. 5 and 6). After the initial cortical incision, little if any bleeding is encountered and neither the electrocautery nor fibrin foam is necessary. The plane of the completed undercutting is then marked by inserting on the nondominant side a loop of stainless steel 30-gauge wire in order to outline the periphery of the undercut area. The ends are bent backward on themselves in order to blunt them for safety's sake. Postoperative lateral and p-a stereoscopic films are taken to show these marking wires (Fig. 7). Saline is then injected between the cortex and dura in order to tamponade the cortex against the undercut area. The dura is closed loosely, bone buttons reinserted without wiring and the scalp sutured with two layers of interrupted silk.

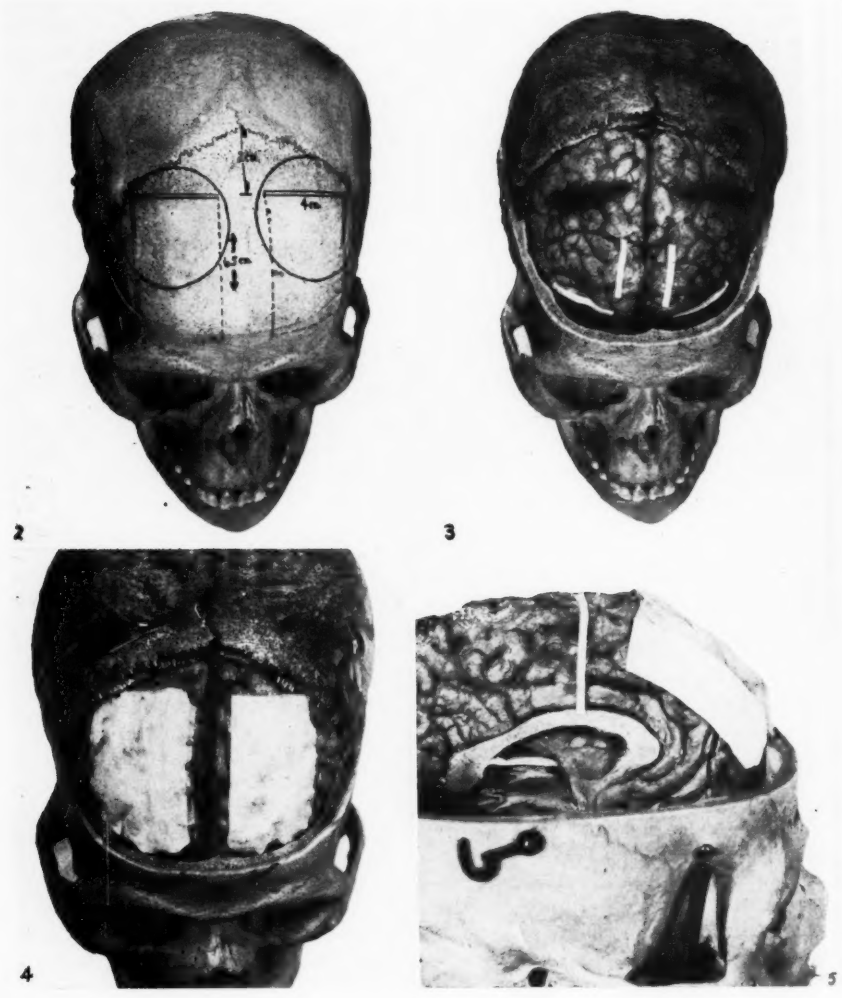

FIG. 2. Location of trephines in undercutting of Brodmann's areas 9 and 10. Solid lines indicate dural incisions. Dotted lines indicate area of undercutting.

FIG. 3. Postmortem brain showing cortical incisions used in undercutting of partial Brodmann's areas 9 and 10. White strips mark the location of cortical incisions as used in orbital and cingulate gyrus undercuttings.

FIGS. 4 and 5. Undercutting of Brodmann's areas 9 and 10 over superior convexity and medial surface.

II. CORTICAL UNDERCUTTING OF THE ORBITAL SURFACE

With the table low and the patient's head moderately extended, a coronal scalp incision is made following the anterior hairline, and the frontal skull exposed subperiosteally down to the orbital ridges. Bilateral 3.75 cm. ($1\frac{1}{2}''$) trephines are placed well lateral to the frontal sinuses with their inferior rims at the orbital ridges and their lateral rims at the lateral angles of the frontal bone (Fig. 8). An inferior dural incision is made exposing the anterolateral tips of the frontal poles. They can be elevated easily to expose the sphenoid ridge in its entirety, the anterior clinoids and optic chiasm. (This same approach has been used for pituitary tumors with an excellent exposure and a marked shortening of the operative time.) Horizontal cortical incisions 3 cm. long are made across the base of the frontal tips (Fig. 3). With

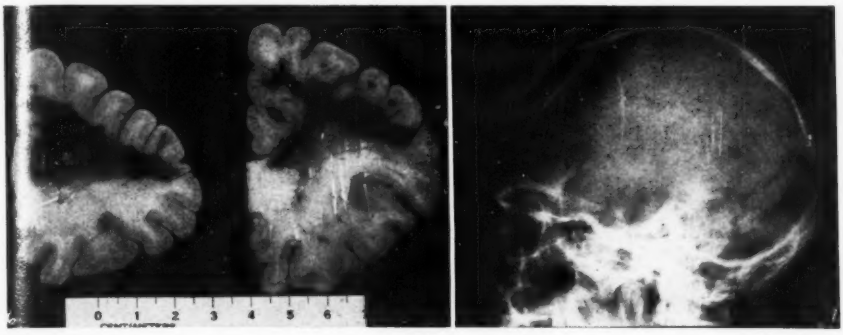

FIG. 6. Vertical cross sections of right frontal lobe demonstrating anterior and posterior levels of undercutting of Brodmann's areas 9 and 10. In this case a more radical undercutting was done for intractable pain, and includes a deeper area extending 1.5 cm. farther laterally and 1 cm. deeper medially than in the 42 other cases reported in this paper.

FIG. 7. Lateral stereoscopic x-ray to show wire markers outlining the undercutting of areas 9 and 10.

spatula-forceps, the line of demarcation of orbital gray and white matter is visualized. The entire orbital surface is undercut including Brodmann's areas 11 and 12 and what constitutes areas 13 and 47 in animals. By inserting the suction tip beneath the frontal lobes to the sphenoid ridge repeatedly and marking with bone wax, one can undercut the orbital surface under direct vision posteriorly to the exact level of the sphenoid ridge, noting that it curves posteriorly as it approaches the clinoid, hence requiring a more posterior undercutting the closer one approaches the midline. Special effort has been made to carry the undercutting to the posterior edge of the anterior clinoids and at this level to angle the undercutting in a basal direction so as to come out at the chiasmatic basal cistern (Fig. 9). This undercutting should include the prechiasmal area. The undercutting is carried laterally and medially to the borders of the orbital surface; a unilateral marking loop of wire is inserted; the dura is closed loosely. Postoperative stereoscopic lateral and p-a x-rays are taken to show the marking wire (Fig. 10).

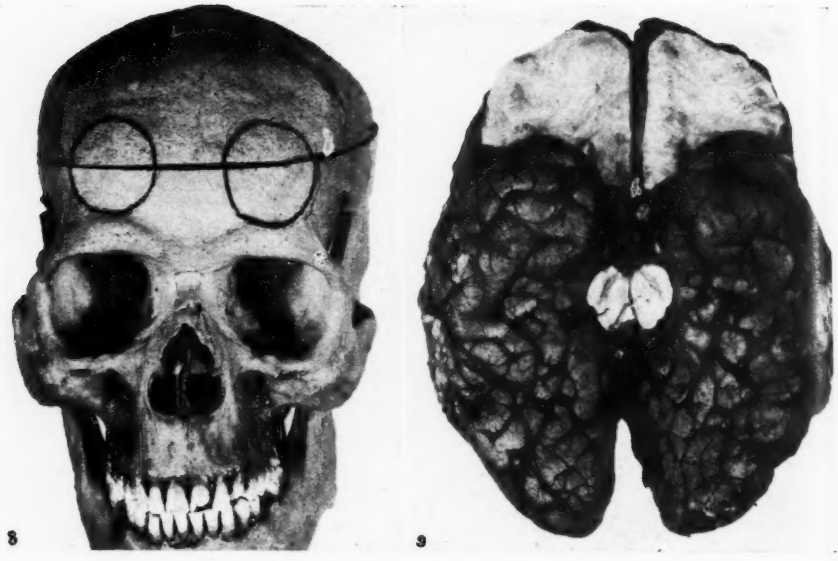

Fig. 8. Location of trephines for undercutting of the orbital surface.
Fig. 9. Orbital undercutting over orbital surface.

Fig. 10. Lateral x-ray to show wire markers in orbital undercutting.

III. CORTICAL UNDERCUTTING OF THE CINGULATE GYRUS AREA

With the table low and the patient's head moderately extended, a coronal anterior hairline scalp incision is made exposing the frontal skull subperiosteally and anteriorly to the frontal sinus. Bilateral 3.75 cm. ($1\frac{1}{2}''$) trephines are made as close to the frontal sinus as possible with their medial edges 5 mm. on either side of the midline (Fig. 11). Straight dural incisions are made parallel to the falx and 1.5 cm. on either side of it. Corresponding cortical incisions are made through the dural incisions. By use of two narrow brain spatulae or special spatula-forceps, the cortex is undercut and separated from the underlying white matter along the medial surface of the frontal lobes back to the corpus callosum. This is identified by extending the undercutting inferiorly and posteriorly to the ependyma of the anterior tips of the frontal horns which lie approximately in the same vertical plane of cross section as the anterior edge of the corpus callosum. The cingulate gyrus surrounding the anterior portion of the corpus callosum is undercut for an approximate length of 3 cm. above the corpus callosum and 2 cm. below it (Fig. 12). Initial attempts had been

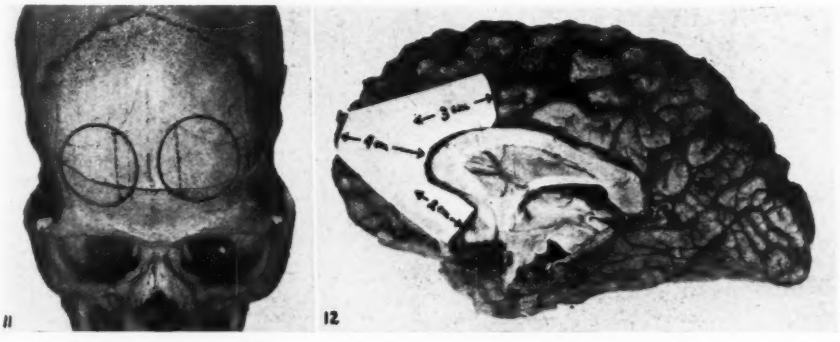

FIG. 11. Location of trephines for undercutting of medial surface and cingulate gyrus.
FIG. 12. Area of undercutting largely limited to anterior cingulate gyrus (area 24) and also area 32 of Brodmann.

made to expose the corpus callosum proper but proved too hazardous because of bleeding and adhesions—hence the use of the anterior tip of the frontal horns of the ventricles as a landmark. Both the genu of the corpus callosum and the anterior tips of the frontal horns lie at an approximate depth of 3.5 to 4.5 cm. from the anterosuperior surface of the brain regardless of the angle of approach. The entire undercutting is carried out by direct vision. Unilateral wire loop markers are then inserted both above and below the ventricle in order to delineate the posterior extent of undercutting of the cingulate gyrus (Fig. 13).

RESULTS

There has been 1 direct operative death due to hematoma, aspiration pneumonia and renal failure and 1 indirect death due to adrenal necrosis, diabetes and invasion of the abdominal viscera in a patient with leiomyo-

sarcoma. Early isolated seizures have occurred in 3 patients, 2 of these having orbital undercutting. One late seizure occurred during a hypoglycemic episode. Anticonvulsants were used only after seizures had occurred. Psychological and mental changes and therapeutic results will be surveyed by the joint hospital staffs at a later date. Physiologic and electroencephalographic results are under study by Zehnder of the Yale Physiological Laboratory and Henry of the Institute of Living. Of research interest are the nega-

FIG. 13. Lateral x-ray to show wire markers extending posteriorly to include the entire anterior cingulate gyrus both above and below the corpus callosum for a distance of 3 cm. from the anterior edge of the genu.

tive physiologic results to date. Few gross physical, visceral or autonomic sequelae have followed even the undercutting of the orbital and cingulate gyrus areas. Prolonged stupor as described by Ward[8] did not occur in those patients undergoing cingulate gyrus undercutting although they were temporarily more somnolent than either the "9 and 10" or "orbital" patients. All were ambulatory and able to eat within 24 hours. Sham rage and maniacal behavior has not followed undercutting of the posterior orbital area. Specificity of frontal lobe function has not yet been demonstrated in our early results. The 3 patients suffering from intractable pain have been initially benefited following cortical undercutting of areas 9 and 10, especially in the relief of anxiety, addiction and anticipation and memory of pain. The duration of relief has not yet been determined.

SUMMARY AND CONCLUSIONS

1. Selective cortical undercutting constitutes a method of producing discrete cortical lesions in man without interference of the blood supply to adjacent areas of the brain.

2. Cortical undercutting is anatomically and surgically feasible and has certain technical advantages over cortical ablations, especially in speed, facility and preservation of blood supply.

3. A preliminary report is made of 43 cases of selective cortical undercutting of the frontal lobes.

4. The areas selected for undercutting have been areas 9 and 10 of Brodmann because of empiric success reported by others in the ablation or leucotomy of these areas, and the orbital and cingulate gyrus areas because of their known influence on the autonomic system and behavior of animals.

5. The operative technique is described, as well as a method of marking the undercut areas by wire loops.

6. The gross physiologic results have been largely of a negative nature.

7. The therapeutic results justify further investigation.

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CASE REPORTS AND TECHNICAL NOTES

MENINGIOMA OF THE FREE MARGIN OF THE CEREBELLAR TENTORIUM

BÉLA HALPERT, M.D., HARRY WILKINS, M.D., AND A. C. LISLE, Jr., M.D.
Department of Pathology and Department of Surgery, University of Oklahoma School of Medicine, Oklahoma City

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The clinical history and the observations at necropsy are presented of a patient who had signs and symptoms of a growth involving the region of the 3rd ventricle. Craniotomy disclosed a seemingly inoperable neoplasm with the microscopic structure of a meningioma. It was discovered at necropsy that the growth originated from the free margin of the cerebellar tentorium near its junction with the falx cerebri and extended forward to reach the 3rd ventricle. The case is reported because of the rarity of a meningioma arising in this location and because of the clinical problem it presented.

REPORT OF CASE

L.B., a white woman aged 20 years, was admitted to the University of Oklahoma Hospitals, Feb. 23, 1946. For about 14 months she had had daily fronto-occipital headaches, with nausea for the last 3 months, but no vomiting. For 2 months there had been blurring of vision with episodes of momentary blindness; impaired hearing, mostly on the right; and difficulty in swallowing; and for 1 month unsteady gait and apraxia of the hands.

FIG. 1. Ventriculogram disclosing asymmetry and dilatation of lateral and 3rd ventricles. The posterior portion of the 3rd is displaced upward and to the right. Superimposed on lower half of left lateral ventricle is a sharply outlined globular field.

Examination. She was oriented, cooperative, alert, with some delay in reaction time. Temperature 98.6°F., pulse 82, and respirations 18. Blood pressure 124 systolic and 82 diastolic. The pupils reacted to light sluggishly, there was moderate papilledema, slight horizontal nystagmus, and right central facial weakness. Gait was unsteady and listed to the right. Babinski and other associated signs were positive on the right. There was loss of ability to perform skilled movements with the right hand. Romberg test was negative, and there was no ataxia and no dysmetria.

Urinalysis was essentially negative. Blood studies: rbc. 4,710,000; hb. 13.5 gm.; wbc. 14,000 with neutrophils 76, lymphocytes 22, and monocytes 2, per cent; Mazzini test negative. Roentgenograms of the head disclosed diffuse thickening of the skull with exaggeration of the convolutional markings, deepening of the sella turcica and thinning of the dorsum sellae. In the posterior parietal region a marked vascularity of the bone was evident. No pertinent changes were seen in a roentgenogram of the chest.

Ventriculograms on March 5 disclosed slight asymmetry and dilatation of the lateral ventricles. A filling defect was noted—a globular structure extending from the posterior portion of the 3rd ventricle upward so that there was some encroachment in the lateral ventricles medially. This was symmetrical on each side (Fig. 1). Ventricular fluid: no globulin; total protein 25 mg./100 cc.; Kolmer-Wassermann test negative; colloidal gold curve 0000000000.

Operation. A right frontal craniotomy disclosed a solid neoplastic mass encroaching upon the left posterior part of the 3rd ventricle. Removal was considered not feasible; a portion was obtained for microscopic study, and the wound was closed.

Histology. The specimen consisted of gray-white tissue, 0.6 by 0.3 by 0.2 cm. Microscopic study (Fig. 4) revealed round or oval, vesicular nuclei in a stream or whorl-like arrangement within a scanty cytoplasm fading into a delicate fibrillar ground substance. In the centers of

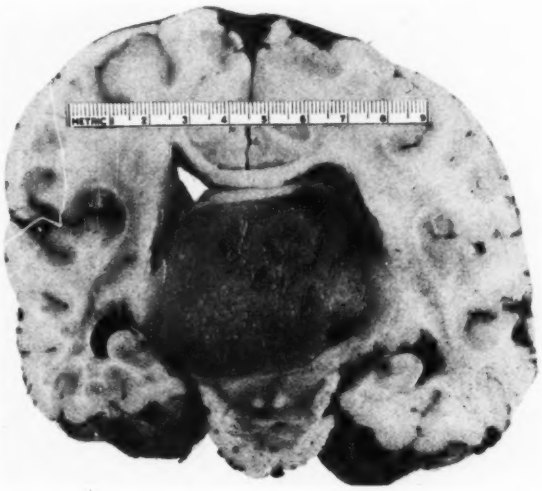

FIG. 2. Frontal section of brain 2 cm. posterior to level of optic chiasm. The tumor occupies the space beneath the corpus callosum and fornix, and between cerebral hemispheres and pons.

some of the whorls there were deposits of lavender-blue granules. In addition to these psammoma bodies there were areas of necrosis and old and more recent extravasations of blood.

Course. The patient's condition became progressively worse, and she died on March 17, 12 days after operation.

Necropsy (12 hours after death) was limited to examination of the cranial content.

The digitations of the inner surface of the calvarium were somewhat exaggerated. The dura mater was tense and over the right frontoparietal region it was covered with organizing clotted blood. Brain protruded through the openings in the parietal regions. The leptomeninx was delicate and transparent. The convolutions were flattened and the sulci were narrowed. No changes were noted in the dural sinuses. The sella turcica was somewhat broadened with the posterior clinoid processes inconspicuous. The hypophysis appeared somewhat flattened.

The brain weighed 1500 gm. The cerebral and cerebellar hemispheres, pons, and medulla oblongata were proportionate and symmetrical. There was a marked pressure cone over the cerebellum up to a depth of 3 cm. A marked pressure groove was noted also on the medial surface of each temporal lobe, about 0.5 cm. outward. The pons was somewhat prominent. The interpeduncular fossa was obscured by bulging of the infundibular region to a height of

1 cm. The brain substance was thinned, obliterating the outlines of the optic chiasm. The vessels of the circle of Willis were delicate and collapsed. Nothing unusual was noted about the cranial nerves.

The cut surfaces of the cerebral hemispheres at the level of the optic chiasm disclosed marked asymmetry of the cavities of the lateral ventricles. There was a shift to the right of the septum pellucidum and of the cavity of the 3rd ventricle. The cavity of the anterior horn of the right ventricle was 1.5 by 2, that of the left 2 by 0.8, and that of the 3rd ventricle, 2 by 0.6 cm. About 2 cm. posterior to the level of the optic chiasm a well circumscribed, globular

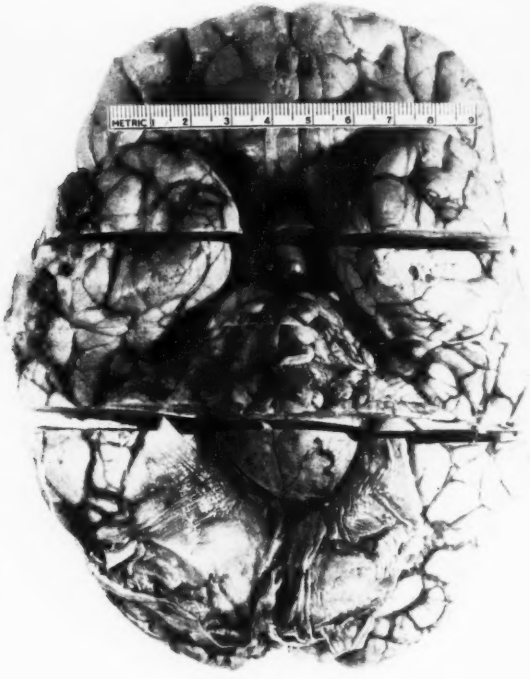

FIG. 3. The attachment of the mass was at the free margin of the cerebellar tentorium on the right of the median line and near the junction with the falx cerebri. The great vein of Galen is on the left side of the mass.

mass, 5 by 4.5 cm., occupied the space beneath the corpus callosum and fornix between the cerebral hemispheres and the pons (Fig. 2). Its cut surfaces had a gray streaked or whorl-like appearance. The mass separated the substance of the cerebral hemispheres, spreading them apart and causing a depression on their medial surfaces as well as on the anterior portion of the vermis cerebelli over an area 2 by 1.5 cm. After removal of the cerebellum the site of attachment of the mass was located at the free margin of the cerebellar tentorium on the right of the median line and near the attachment of the falx cerebri for a length of about 1 cm. The great vein of Galen was located above and to the left of the upper convex surface of the mass (Fig. 3).

Microscopic preparations from various parts of the growth disclosed a structure similar to that seen in the specimen removed at operation (Fig. 4).

COMMENT

When a meningioma originates from the part of the dura mater that lines the cranial surface, the overlying osseous tissue frequently undergoes a change demonstrable by roentgenography. This change consists of a focal thickening with some condensation of the osseous pattern, and, with the other general and localizing signs and symptoms of an intracranial neoplasm, is helpful in determining the exact site of the growth. No such localizing aid is available when the meningioma originates

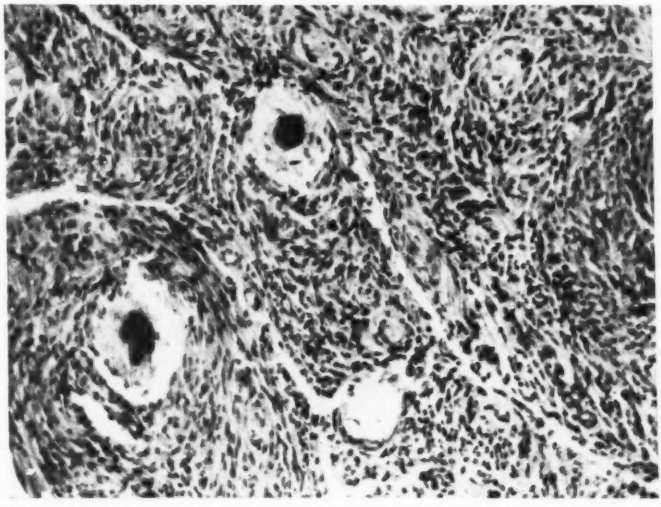

FIG. 4. Microscopic appearance of the growth (described in text). ×150.

from either the falx cerebri or the cerebellar tentorium away from their cranial attachments. In retrospect, perhaps, a postero-anterior view which was not obtained would have given a clue as to the extent and possibly as to the origin of the growth.

The sites of origin of meningiomas have been carefully tabulated by Cushing and Eisenhardt[2] and more recently considered by Olivecrona[4] and Grant.[3] With the exception of the case illustrated by Balado and Tiscornia[1] (cited by Cushing and Eisenhardt) none was claimed to have originated at the same site as ours, namely, the free margin of the cerebellar tentorium near its junction with the falx cerebri.

SUMMARY

A meningioma which produced clinical signs and symptoms of a tumor of the 3rd ventricle is reported. The neoplasm originated from the free margin of the cerebellar tentorium on the right near its junction with the falx cerebri and extended forward beneath the corpus callosum to reach the 3rd ventricle. This is believed to be the 2nd such case ever recorded.

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CEREBELLAR EXTRADURAL HEMATOMA

REPORT OF A CASE

Arthur Bacon, M.D.*
Wichita, Kansas

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The literature contains 3 reports of extradural hematoma in the posterior fossa treated by operation.[1,2,5] All 3 patients recovered. Fifteen additional cases, perhaps more, have been reported from autopsy material.[3,4,6] Adequate clinical observation, however, is so scarce that an easily recognizable syndrome for this lesion can not yet be described. In some, it appears that the progress of neurological signs extends over a period of a week or more, allowing for relatively simple localization of the lesion. In others, the progress is rapid and no localizing features are observed until compression of the cerebellum and brain stem is far advanced. Unless relieved by prompt operation, the patient may then expire following a short interval of coma and hypotonia of all extremities.

The following case of cerebellar extradural hematoma is an example of the type that pursues a less emergent course.

CASE REPORT

A 27-year-old oil field laborer was admitted to the Veterans Hospital of Wichita on Aug. 31, 1947, for treatment of a head injury of 8 days' duration. His past history was not relevant.

On Aug. 22, 1947, he fell from a doorstep, striking the back of his head on the sidewalk. He was unconscious for 15 minutes, then recovered consciousness and was taken home. For the following 8 days he was confined to bed by headache, vertigo, nausea and vomiting. It could not be ascertained whether his state of consciousness fluctuated, improved or became progressively worse, since the patient was an unreliable witness in regard to this symptom. However, a mild degree of drowsiness was present on admission, and increased moderately while under observation.

General physical examination was not unusual. Pulse was 88, and blood pressure 135/90. There was nystagmus and papilledema. Headache was constant and he ate poorly because of nausea. On September 2, a lumbar puncture was performed. Xanthochromic spinal fluid was obtained under an initial pressure of 500 mm. of water. This was reduced to 220 mm. by removal of 7 cc. of fluid. Queckenstedt test: no evidence of subarachnoid block. Neurosurgical consultation was then requested.

Examination on September 2, 11 days after injury, elicited the following: The patient was drowsy but easily aroused. He complained of frontal headache. Speech was slurred and had a

* 203 KFH Building, Wichita, Kansas.

nasal quality. The neck was slightly stiff. There was contusion of the scalp 1 inch to the left of the inion. The patient preferred to lie on his side. Change of position consistently produced vertigo, nausea and a slow horizontal nystagmus of short duration. On attempting to fix his gaze straight ahead, the eyes would deviate slowly to the right. Pupils were equal and reacted well to light and near vision. External ocular movements were not restricted. There was mild papilledema bilaterally. No visual field defect could be demonstrated. There was complete paralysis of the left soft palate. Except for mild central paresis of the left side of the face, the remainder of the cranial nerve functions were normal.

The outstanding localizing abnormalities were cerebellar signs involving the left extremities. Adiadokokinesis, dysmetria, decomposition of movement, hypotonia and absence of check were found in the left upper extremity, and ataxia in the left heel-to-shin test. Tendon reflexes were more active on the left. Plantar reflexes were normal. There was no disturbance of pain perception, position sense, stereognosis or vibration perception. Roentgenograms of the skull failed to reveal a fracture owing to the omission of an occipital view. The clinical impression was hematoma in the left posterior fossa, with probable contusion of the right temporal lobe.

Operation, Sept. 2, 1947. A left suboccipital exposure was made, disclosing a fissure fracture extending diagonally across the left half of the occipital bone. A craniotomy over the center of the cerebellar hemisphere revealed an extradural clot of currant-jelly consistency confined to the left side of the posterior fossa. The clot was 2.5 cm. in thickness in a circumscribed area directly beneath the fracture line, but diminished rapidly in thickness toward the periphery, so that the total volume of the clot was estimated to be not more than 15 gm. Upon removal of the clot a laceration of the dura, 3 cm. in length, was exposed coinciding with the line of fracture, and at this point lacerated cerebellar cortex was visible. The cerebellar hemisphere was explored without encountering an intracerebellar hematoma. The dura re-expanded very slightly during the procedure. Bleeding points along the dural laceration were controlled without difficulty and the wound was closed without drainage.

The patient improved steadily following operation and was discharged on the 16th postoperative day. Examination on October 20, 38 days following removal of the hematoma, revealed no neurological abnormalities. The patient was free from symptoms and ready to resume work.

COMMENT

In this case, diagnosis of hematoma in the left posterior fossa was not difficult. The history of a moderately severe head injury followed by the appearance of papilledema, nystagmus, incoordination on the left, and paralysis of the left 10th cranial nerve directed attention inevitably to the left posterior fossa. No attempt was made preoperatively to predict the location of the hematoma, as to whether it was extradural, subdural or intracerebellar. The extensive functional loss appeared to favor the intracerebellar location. The patient had, however, been subjected to a Queckenstedt test and had withstood reduction of intracranial pressure by withdrawal of spinal fluid, with no ill effects. The fact that he did not succumb to these studies was thought to favor the extradural location.

The writer believes, with many others, that the Queckenstedt test should be carefully avoided in the diagnosis of intracranial lesions. It is of interest, however, to note that Kessel's case[2] of cerebellar extradural hematoma did show a complete block by the Queckenstedt test.

The majority of the reported instances of cerebellar extradural hematoma have been encountered at autopsy. LeCount and Apfelbach[3] reported 8 cases among 504 autopsies involving skull fracture. Vance[6] encountered 4 cases in a series of 512 autopsies, also involving skull fracture. Both these authors describe the lesion as arising from lacerations of the transverse sinuses by fracture. In some of these, the

hematoma extended above the transverse sinus as well as downward into the posterior fossa. It seems plain, however, that cerebellar extradural hematoma may arise from sources other than the transverse sinuses, and further, that a fracture need not be present. In the case here reported, it seems probable that the hematoma resulted from torn vessels associated with laceration of the dura which did not extend into a dural sinus. In the case reported by Turnbull[5] there was no evidence of fracture by x-ray or at operation. Turnbull made no comment relative to the source of the original hemorrhage, but described the hematoma as lying well away from the transverse sinus.

The writer found only 4 reports of this lesion accompanied by adequate clinical observations. In 1 of these (Turnbull) the progression of neurological signs over a period of 3 weeks led to the preoperative diagnosis of cerebellar tumor. The patient recovered uneventfully following the removal of a unilateral cerebellar extradural hematoma. In this patient, as in the 1 here reported, the correct surgical approach was not difficult to determine.

In the remaining 3 cases, however, the courses were rapid and the neurological pictures were entirely different. This rapid type of clinical course represented a surgical emergency and allowed little time for diagnostic procedures. Because of the similarity in these cases it seems worthwhile to summarize them briefly.

McKenzie's[4] case was that of a child who suffered a mild head injury, became stuporous after a lapse of 30 hours, had one tonic convulsion and expired 14 hours after the onset of the stupor. The only neurological abnormality was a striking flaccidity of all extremities. Necropsy showed an extradural clot limited to the right posterior fossa. There was a fracture of the occipital bone.

The case of Coleman and Thomson[1] also presented a rapid course. A child of 9 fell and struck the back of her head, but was not unconscious. Neurological examination was negative. Twenty-four hours later headache, drowsiness and vomiting began, but again neurological examination failed to reveal any abnormality. A few hours later, however, stiff neck, hypotonia of all extremities and areflexia were found. The patient recovered following the removal of a bilateral cerebellar extradural hematoma.

Kessel's case[2] was that of a 24-year-old woman who fell, striking the back of her head. She was unconscious a few minutes and complained thereafter of severe headache. Roentgenograms showed a slight separation of the left lambdoid suture. Thirty hours after injury she had a tonic convulsion and the next day passed into coma following two more tonic episodes. There was no hypotonia or diminution of reflexes. The Queckenstedt test showed complete subarachnoid block. At operation a left cerebellar extradural clot was removed. The patient recovered.

No attempt to describe a syndrome is warranted from this scanty material. It would seem, nevertheless, that stupor, with hypotonia, or tonic convulsions, appearing after a lucid interval of 24 hours following injury, should bring to mind the possibility of cerebellar extradural hematoma.

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MENINGEAL MENINGIOMATOSIS

REPORT OF CASE

Alfred Uihlein, M.D., Edward M. Gates, M.D.,* and Robert G. Fisher, M.D.*
Section on Neurosurgery, Mayo Clinic and Mayo Foundation, Rochester, Minnesota

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In 2,023 cases of intracranial tumor, Cushing,[10] according to his own classification, described 271 cases of meningioma representing 13.4 per cent of his total cases. Baker,[5] in 757 cases of intracranial tumor found 122 cases of meningioma, or a percentage of 16.1, while Courville[9] found, at autopsy, 71 meningiomas in 561 intracranial tumors, an incidence of 12.7 per cent. This type of tumor, for the most part, represents a benign lesion and frequently is operable. However, it is important from both an academic and surgical viewpoint to realize that this type of lesion may occur in multiple sites; a number of case reports of multiple meningiomas have appeared in the literature in the last fifty years.[1,2,7,12,15,16,17,19,21,22] Frequently, a number of tumors are found in such conditions as von Recklinghausen's disease with no explicable cause. Cases of multiple meningiomas, according to the literature, are comparatively rare. The case to be reported represents one that we have elected to classify as "meningeal meningiomatosis."

REPORT OF CASE

The patient was a 2½-year-old boy who had had a normal birth and development. About April 1, 1947, he complained of episodes of backache in the sacral region. The mother noted that when she picked the child up by grasping him under the arms he would cry out with pain. These pains and the backache occurred periodically each day but he continued to play normally. However, he would awaken at night screaming with pain, which lasted but a short time, and then he would fall asleep.

In May, 1947, the mother noticed that the boy began to walk "like a little old man who needed a cane." He complained of pains in the right knee and thigh, and would awaken from his sleep nearly every 2 hours during the night crying with pain in the back and leg. He would not bend the right leg or thigh beyond an angle of 45° because of the severe pain that such motion caused him. Roentgen studies of the spinal column conducted by the family physician were reported as negative.

In June, 1947, the patient was no longer able to walk upright and had to crawl to get about his home. He was hospitalized at that time in the vicinity of his home. Repeated lumbar punctures were attempted but fluid was not obtained. Fluid obtained on cisternal puncture was reported to have contained 15 cells/cc., with 20 per cent polymorphonuclear leukocytes and 80 per cent lymphocytes; protein 50 mg./100 cc.; globulin test positive; cultures failed to show any growth of organisms. While the patient was in the hospital, there developed right facial paralysis and weakness of the right leg which was followed in a few days by weakness of

* Fellow in Neurosurgery, Mayo Foundation, Rochester, Minnesota.

the left leg. He experienced considerable pain in his back and legs at that time. Passive motion of his legs elicited rather severe pain. A diagnosis of brain tumor was made and the patient was brought to the clinic on July 4, 1947.

Examination. The boy was lying in bed crying with pain. He was normally developed and well nourished. Temperature was 99.8°F. There was ptosis of the right upper lid. Questionable nystagmus was noted on left lateral gaze. Ocular movements were normal. The pupils were equal and their responses to light and convergence were normal. His speech did not show any abnormalities. There was complete paralysis of the muscles of facial expression on the right side. Motor power was normal in the left upper extremity and left side of the trunk. The muscles of the right upper extremity and right half of the abdomen were weaker than normal. There was definite weakness of the muscles of the right lower extremity; the muscles of the left lower extremity were weak but the weakness was not as great as that of the right. The triceps reflexes of both arms were absent. The quadriceps surae and triceps surae reflexes were bilaterally absent. No Hoffmann or Babinski reflexes were obtained. Kernig's sign and Lasègue's sign were present in both extremities. A moderately stiff neck was noted. The anal sphincter was completely relaxed and paralyzed. The boy was incontinent of both feces and urine.

Sensory examination gave normal results except for complete loss of all modalities in the region of the buttocks, perineum and posterior and anterior aspects of the thighs. Pin prick over the legs produced intense pain characteristic of hyperesthesia. Funduscopic examination did not disclose any papilledema or lesions of the retina. The results of laboratory studies of the urine and blood were essentially normal. Flocculation studies for syphilis gave normal results. Roentgenograms of the entire spinal column, head and thorax failed to show any abnormalities. Lumbar punctures were attempted on several occasions but were always unsuccessful since no spinal fluid could be obtained.

An EEG made on July 9, 1947, revealed generalized delta activity that was most marked posteriorly. At that time deafness in the right ear was noted. The diagnosis of intracranial neoplasm with metastasis to the lower part of the lumbosacral region was made.

Operation. Because of the severe and unbearable pain which could not be relieved by medication, a lower dorsolumbar laminectomy with decompression was carried out by one of us (A.U.) on July 17, 1947. During the operation it was noted that the filum terminale was consolidated into one large tumor mass. The specimen taken at this time was reported by the surgical pathologist as being a highly malignant tumor, possibly a malignant meningioma.

Postoperative course was satisfactory and the patient was relatively comfortable. Since it was felt that this type of tumor might respond to roentgen therapy, a palliative course of treatment was given to the cerebellar region. However, the disease continued to progress in spite of this and evidence of acute increased intracranial pressure became apparent. In order to relieve this acute situation a ventriculostomy with continuous ventricular drainage was performed. The boy remained comfortable until the final day of his illness when convulsions developed; death occurred on the 36th hospital day.

Necropsy. A careful examination of all the body tissues and organs was made to determine whether the neoplasm of the central nervous system was a metastatic or a primary growth. No evidence of metastatic lesions could be found anywhere outside of the central nervous system. The brain and spinal cord weighed 1,175 gm. (Fig. 1). There was a well-healed scar, 8 cm. long, over the spinous processes in the midline extending from the level of the 9th thoracic spine to the level of the 1st sacral vertebra. A small right occipital trephine opening where a ventricular catheter had previously been in place was noted. The calvarium over its entire extent was unusually thin.

The dura was tense and adherent to the skull in the occipital region around the foramen magnum, where there were multiple soft, purplish-colored pedunculated tumors. In the region of the right petrous bone surrounding the internal auditory foramen and growing into the foramen, a flesh-colored soft pedunculated mass engulfed the auditory and facial nerves. A similar condition obtained about the left auditory foramen, auditory nerve and facial nerve. About the posterior clinoid region extending to the anterior border of the foramen magnum

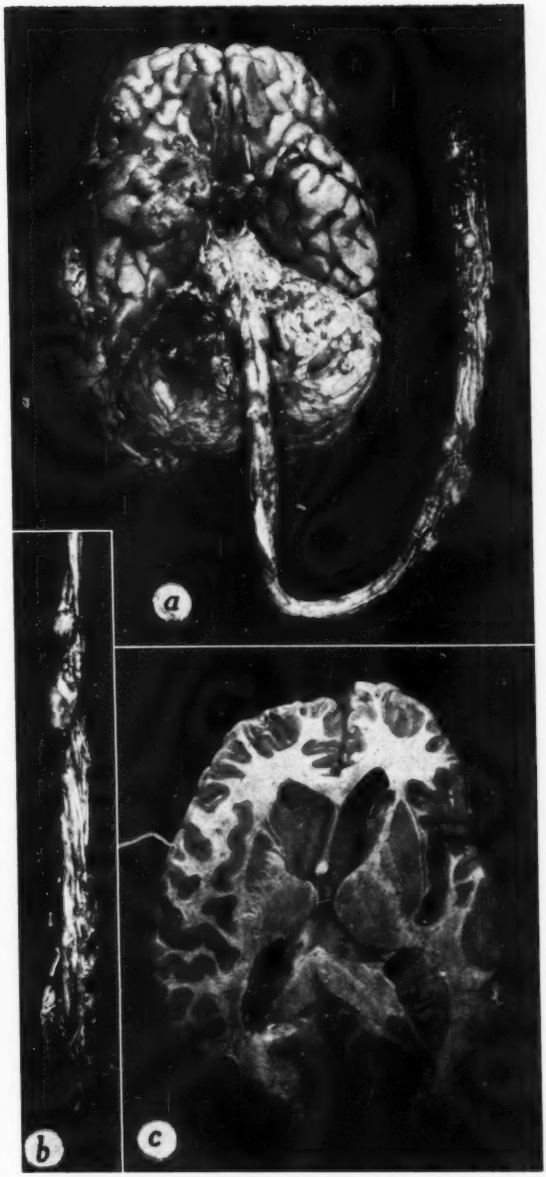

FIG. 1. *a.* Gross appearance of the brain and spinal cord. *b.* Many tumors are present along the course of the cord. *c.* Horizontal section through the brain.

another soft flesh-colored pedunculated mass was noted. The venous sinuses were all unobstructed.

The arachnoid over the brain and cerebellum was thin, delicate and translucent except in multiple areas over the lobes of both cerebellar hemispheres. In these small areas the lesion appeared slightly elevated, opaque and pedunculated. The arachnoid throughout the entire length of the spinal cord and the filum terminale was surrounded by tumor growth. The growth consisted of multiple pedunculated masses which had grown freely from anterior and posterior nerve roots into the cord substance, forming thick plaques encircling the cord. The filum terminale was a solid mass of tumor tissue.

Examination of the cerebrum disclosed small fleshy pink tumors in the choroid plexuses of both anterior and posterior horns of the lateral ventricles. The ventricular system was slightly dilated. A small pink friable mass had infiltrated the posterior limit of the right side of the thalamus. The 3rd ventricle was anatomically open. The 4th ventricle contained a pedunculated flesh-colored tumor which almost completely filled the chamber (Fig. 2*a*). The cerebellopontine angles were free of tumor tissue. The right anterior inferior surface of the cerebellum was deeply grooved by a pedunculated friable neoplastic mass.

Multiple cross sections of the brain stem revealed tumor tissue that had grown into the anterior portion of the pons and medulla. The spinal cord, on cross section, at no level was found free of neoplasm (Fig. 2*b*, *c*, *d* and *e*). In the lumbar region the cord was compressed and infiltrated to the extent that nothing remained but the posterior horns of gray matter and the posterior fasciculi. The filum terminale was a solid sheet of tumor which surrounded and compressed the nerves.

Microscopic Examination. The dural masses were found to be composed of a dense collagenous stroma in which were imbedded islands of oval-shaped, pale, pink-staining cells with elliptical nuclei. The cells were highly pleomorphic and grew in no characteristic pattern. Tumor giant cells were present but not in any great numbers. In some sections, the nuclei of these neoplastic cells were hyperchromatic and showed abnormal mitotic figures. In general, a fair amount of chromatin material was present about the periphery of the nuclei. The tissue was well vascularized, with the vessels running in the collagen network. Some adventitial and intimal proliferation could be found in the network of vessels.

In routine sections of the cerebral lobes in which tumor tissue was not suspected grossly, such tissue was found microscopically. These sections showed the masses to be arising from the arachnoid cap cells and streaming off along the pia-arachnoid down deep into the sulci and infiltrating the cortex (Figs. 3 and 4). This was a common picture seen throughout the entire cerebrum and in the cerebellum. The picture was again repeated when sections of the pons and medulla were examined. Microscopic study of the cerebellum revealed small bits of tumor growing between the folia and into the cerebellar tissues. The choroid plexuses were engulfed in neoplastic cells.

Sections of the spinal cord showed the same type of tissue and the same ingrowth of tumor tissue from the pia-arachnoid. The picture was consistent with that of highly malignant meningiomatosis of meningeal origin. No primary neoplastic site could be found. It was felt from studying these sections that the entire arachnoid had, in various and multiple locations, undergone malignant changes, giving rise to many primary malignant tumors which were highly invasive. The term "sarcoma of the meninges" was avoided for it was felt that insufficient evidence exists to justify classification of the meningiomas as of mesodermal origin. Thus until such time as it could be shown that the meninges arise from mesoderm, it was felt better to adhere to a different term for these arachnoid tumors; accordingly, in this case the term "malignant meningeal meningiomatosis" was used to describe the condition.

The tissues were seen by Dr. J. W. Kernohan who concurred in the diagnosis and in the justification for the term "meningeal meningiomatosis."

COMMENT

In the preoperative study of this patient, the clinical diagnosis was intracranial tumor; it was thought that this tumor was the primary source for a metastatic lesion

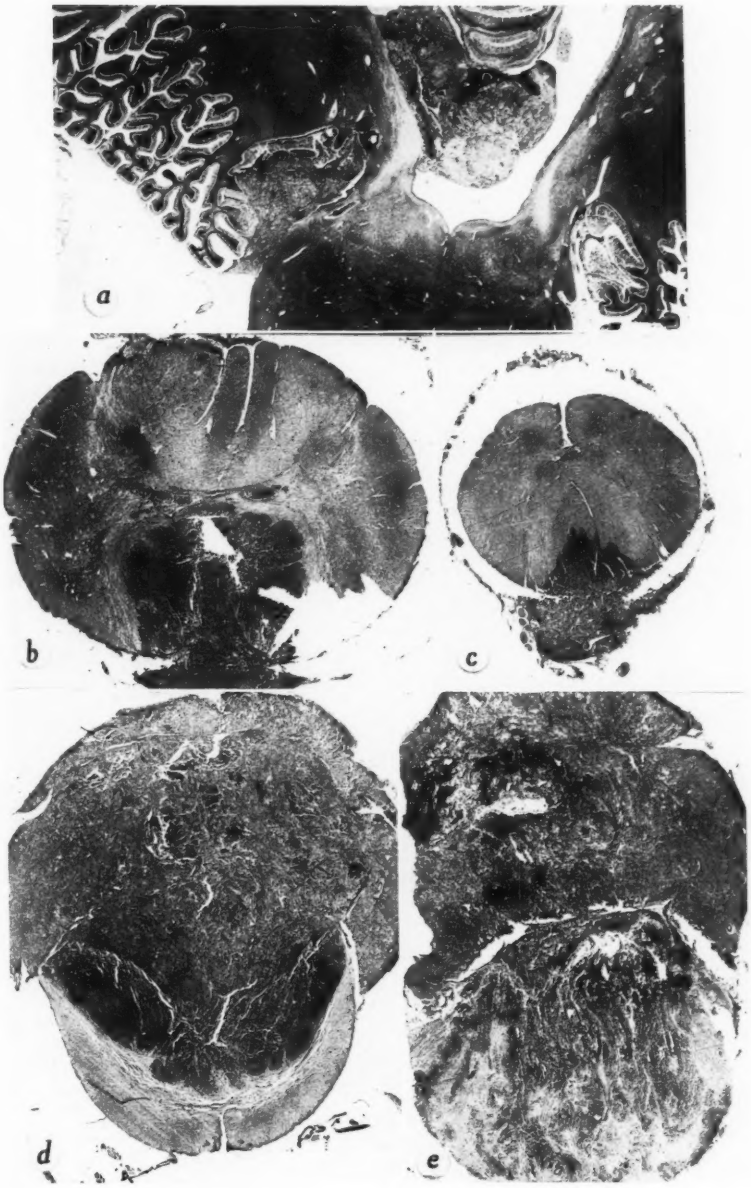

FIG. 2. Sites where tumor tissue was found. *a*. Fourth ventricle (×2½). *b*. Cervical cord (×5½). *c*. Thoracic cord (×5½). *d*. Lumbar cord (×5½). *e*. Filum terminale (×5½).

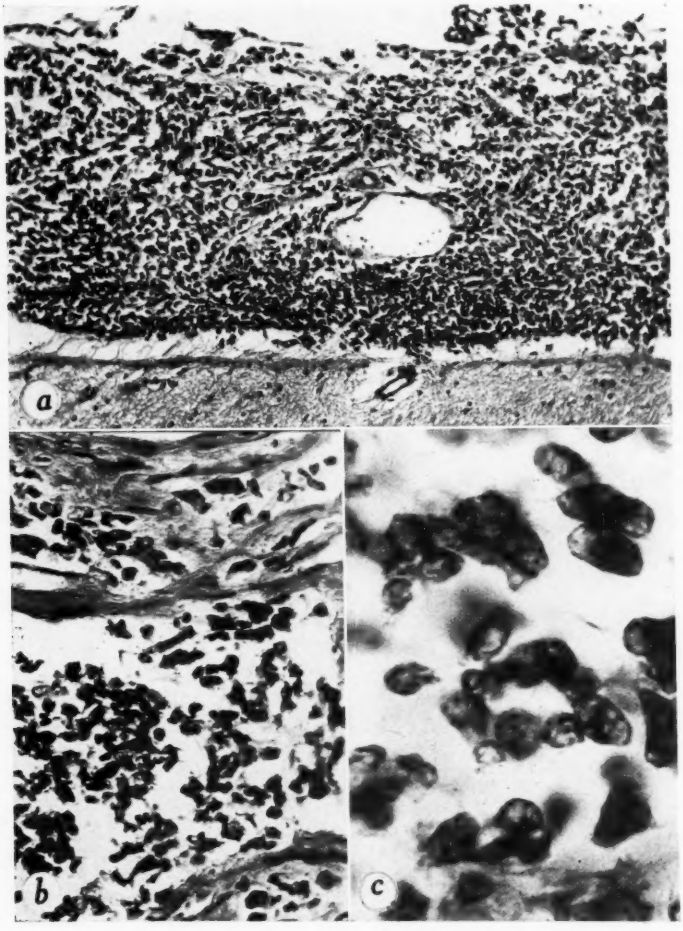

FIG. 3. Site of origin of tumor tissue from the arachnoid. *a*. ×95. *b*. ×265. *c*. ×1,000.

in the spinal column. The degree of malignancy was determined at the time of the exploratory laminectomy but the degree of involvement of the nervous system by such a diffuse process was not determined until the postmortem examination had been completed. The pathologic classification of a case of this type is difficult and again brings up the question of the origin of the meninges. If one assumes that the meninges are of mesodermal origin, then the findings in this case might represent an instance of primary sarcoma in the meninges, with secondary metastasis to other parts of the leptomeninges. The other assumption is that this case represents diffuse transition of normal meningeal tissue to neoplastic tissue.

Schultze,[20] in 1880, reported a case of "multiple pial sarcoma" and Nonne,[18] in 1902, substantiated this idea and described tumor cells in the pial vessels which secondarily invaded the meninges. Fried[13] described a rare condition under the title of "diffuse sarcomatosis of the meninges" in which there had been a general change in the nature of the leptomeninges. Bailey,[3] in 1929, stated that the meninges were of neuro-ectodermal origin but recognized that intracranial sarcomatous tumors

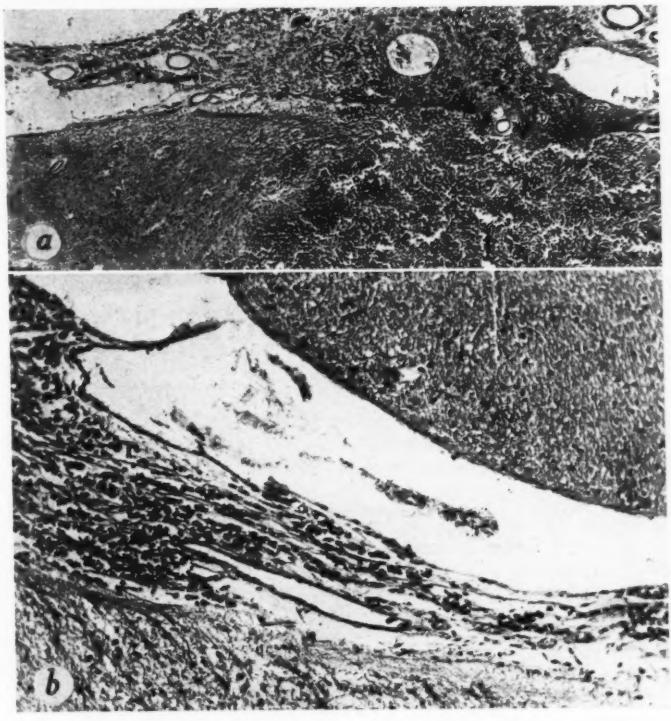

FIG. 4. *a*. Tumor cells growing from the arachnoid into the cortex (×95) and, *b*, into the convolutions (×95).

may have leptomeningeal origin. Bailey and Bucy,[4] in discussing the origin of meningeal tumors, stated that tumors such as we have described are of connective-tissue, not gliomatous, origin. These authors assumed that meningeal mesenchyme derived from neuro-ectoderm or mesoderm might give rise to the various types of meningioma.

Globus, Levin and Sheps[14] stated that the pia is of mesodermal origin and is derived from a primitive endomeninx. Also they said that the endomeninx, with the ectomeninx which is the periosteum, and the dura had a common precursor in the skeletoneural intertissue which was derived from the mesenchyme. Cushing and Eisenhardt[11] stated that the term "multiple meningiomas" referred to several dis-

crete tumors, but that if the neoplastic nodules were numerous and coalesced, then the condition was meningiomatosis or diffuse meningioma. Previously Connor and Cushing[8] cited reports of 2 cases in which diffuse tumors of the meninges were apparent only on microscopic examination and the condition in these cases was called "true sarcomatosis" or "endotheliomatosis." It was their impression that the tumors originated in the adventitia or perivascular sheaths of the cortical blood vessels. Brown and Kernohan,[6] in 1941, limited the term "meningiomatosis" to an arachnoid type of lesion and assigned purely mesodermal tumors to the category of sarcoma. In their case, the cells had morphologically benign characteristics. They reasoned that the process of meningiomatosis resembled a neoplastic transformation of multicentric origin arising on an incompletely differentiated arachnoidal membrane. Since there is no further evidence to justify classification of the histopathologic features seen in our case differently, we have used the title "meningeal meningiomatosis."

SUMMARY

The case of a $2\frac{1}{2}$-year-old boy is reported in which a diffuse transitional process in the meninges resulted in multiple tumors of the leptomeninges with malignant characteristics. The term "meningeal meningiomatosis" is used to describe the pathologic characteristics observed.

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THIRD VENTRICULOSTOMY PROVEN PATENT AFTER FIFTEEN YEARS

Ira Cohen, M.D.*
New York, N. Y.

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When obstructive hydrocephalus in adults is due to a lesion that is not removable, one of two palliative procedures may be employed. The obstruction may be bypassed by a tube leading from the lateral ventricle into the cisterna magna, as described by Torkildsen.[5] Or the accumulation of excess fluid in the ventricle may be prevented by opening a communication between the 3rd ventricle and the basal cisternae. This procedure, first described by Dandy[1] in 1922, was devised for congenital hydrocephalus in older age groups. Among others its use has been described by Stookey and Scarff[3] and by White and Michelsen.[6] The latter reported on its use in 11 cases; of these, the longest survival up to the time of reporting was $2\frac{1}{2}$ years. In Dandy's report in 1945[2] there was 1 patient living $23\frac{1}{2}$ years after operation and 3 living between 10 and 20 years.

It is a fair assumption that these long survival periods are dependent on the continuous patency and functioning of the artificial stoma, though the verification of such a conclusion is lacking. In a case reported by Sweet[4] the spontaneous ventriculostomy found at necropsy had, to judge by the history, functioned for 12 years. The following case report yields additional proof of the long-range patency of a ventriculostomy.

CASE REPORT

In 1931, a young woman aged 17 years was admitted to the Neurological service of the Mount Sinai Hospital, complaining of adiposity of 2 years' duration and visual difficulty for 4 months. She had always been plump and became definitely stouter with increase in food consumption. The first visual difficulty was that of black spots before both eyes. Then vision in the left eye became impaired, followed by loss in the right eye. Two months prior to admission vision was so poor that she was unable to distinguish objects or colors. For 2 months she had complained of tinnitus in the left ear with some loss of hearing in both ears, slight vertigo and occasional headache. There was nothing in the history to suggest polyuria, polydypsia, nor menstrual disturbances.

Examination. She was noted to grope her way about the ward because of visual loss. Her

* 4 East 95th St., New York 28, N. Y.

extremities were large proximally and tapered distally. Her breasts were very large. There was right-sided hyperreflexia, slight bilateral exophthalmos and inconstant nystagmus in extreme lateral gaze. Papilledema of $2\frac{1}{2}$–3 D. with pale discs suggesting atrophy was present. The skull plates showed some enlargement of the sella with destruction of posterior clinoids and partial loss of dorsum sellae. Vision was too poor to do perimetric studies.

The cerebrospinal fluid was under a pressure of 340 mm. water; total protein content was 30 mg. per cent.

Because of the enlarged sella, loss of vision, and the endocrine disturbances operation was undertaken for a probable pituitary tumor.

Operation. Nov. 27, 1931. A right frontal bone flap was fashioned. When the anterior horn of the ventricle was tapped it was found to be dilated. Some 60 cc. of fluid were withdrawn.

FIG. 1. Calcified mass.

Behind the chiasm a bluish cystic mass was exposed. It yielded blood-tinged clear fluid. Its thin wall was torn into and large amounts of clear fluid welled up into the field, more than could be accounted for by the size of the "cyst" exposed. It was assumed that the cyst was a multilocular one, that it extended backward and that it pressed upward into the 3rd ventricle, causing the hydrocephalus.

Postoperative course was gratifying, and vision improved steadily. Six weeks after operation visual fields showed a binasal hemianopsia.

The follow-up notes showed at 3 months she counted fingers at 8 feet.

In 1933 she reported that a thyroid adenoma had been removed.

In 1934 she could read large print.

In 1937 she reported she had had a pregnancy interrupted and that she had lost 50 pounds in weight.

She failed to report again until 1946. She was referred back to the hospital with com-

plaints of amenorrhea for a year, diminished hearing for 2 or 3 years and a recent gain in weight. There was also an unacknowledged diminution in vision. She complained of occasional headache.

On examination there was a paucity of abnormal signs. She was obese. Deafness of a conduction type was found bilaterally. Vision in the right eye was 20/70, in the left 20/200. The fields suggested a binasal hemianopsia. The fundi showed bilateral atrophy. Spinal puncture yielded clear fluid under a pressure of 140 mm. water. The total protein content was 30 mg. per cent. The basal metabolic rate was −21. Roentgenograms of skull (Fig. 1) showed a normal sella and a calcified mass in the midline (which was subsequently shown to be in the posterior fossa).

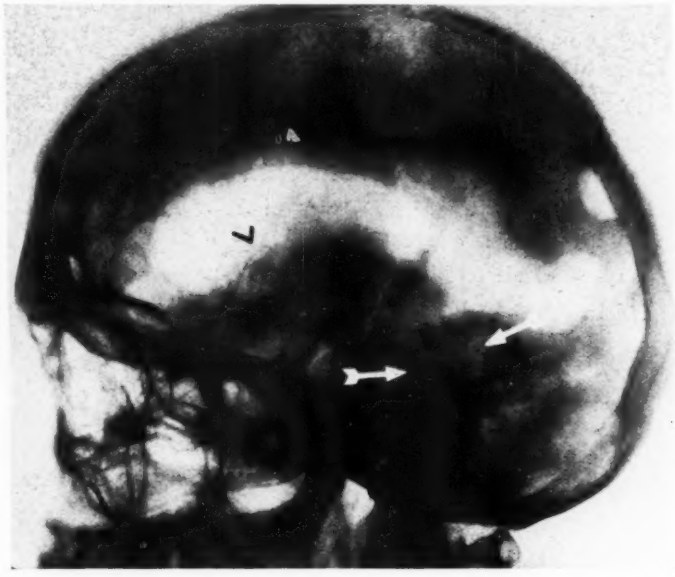

FIG. 2. Ventriculogram showing hydrocephalus and relation of calcification to the ventricles.

A pneumoencephalogram was attempted but no air entered the ventricles. The first ventriculogram did not supply all the information desired, so another was done 3 days later. Again dilated ventricles were found but the 3rd did not visualize (Fig. 2). The patient was not operated upon immediately. She went into stupor the following day and was then operated upon.

April 13, 1946 a suboccipital craniotomy was performed. Before opening the dura the arch of the atlas was removed. The cisterna magna was distended with fluid. The cerebellar tonsils were not herniated. In the upper portion of the vermis a calcified tumor was encountered. Only part of it could be removed. The patient remained in stupor and died on the 4th postoperative day.

Autopsy. The essential findings were reported by Dr. J. H. Globus as follows: "The brain is of average size. The surface appearance reveals nothing unusual except for a large defect in the anterior portion of the interpeduncular space (Fig. 3). This defect measures about 1 cm. in diameter and communicates the interpeduncular space with the interior of the 3rd

ventricle. Its posterior boundary is the optic chiasm; anteriorly it has almost completely destroyed the exposed part of the lamina terminalis. The borders of the defect are rounded and somewhat thickened. The vermis of the cerebellum is almost completely replaced by an area of disorganized and discolored tissue. This area of disorganization and discoloration involves the adjacent portion of the cerebellar hemisphere. This apparently was the site from which tumor tissue was removed at operation.

"On making a middle sagittal section (Fig. 4) a large area of disorganization is found in

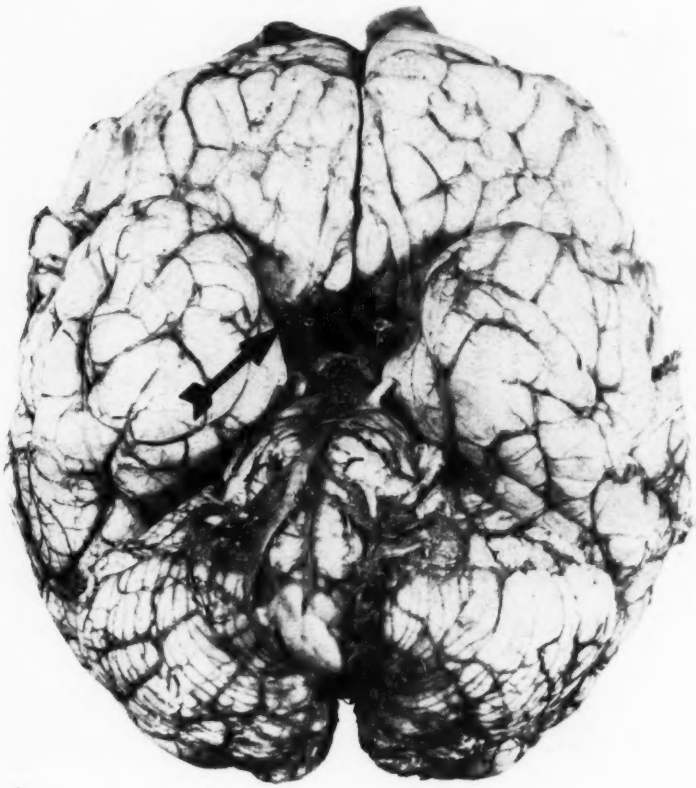

FIG. 3. Patent stoma of ventriculostomy.

the cerebellum, affecting the vermis and adjacent portions of the hemispheres. At the midline there is only a narrow zone of cortex and vermis left. In the brain stem just above the pons and extending a short distance in the pons there is also a fairly large area of disorganization. The aqueduct is represented by a fairly large mass of hard tissues measuring about 2 cm. in long plane and 2½ in vertical plane. The pineal body is triangularly flattened, apparently from compression by the tumor mass below and the splenium of the corpus callosum above.

Microscopic Descriptions. "Section of the brain adjacent to the surgical defect shows a wide zone of tissue which consists mainly of interlacing bundles of elongated cells. Near the free

border of this layer are many psammoma bodies and small deposits of calcified material. Here and there are small vascular channels. In another area the vascular channels are very prominent and the psammoma bodies are better organized. Cajal preparations show that the streaming elongated cells are in all probability of glial origin. Only an occasional cell is, however, an outspoken astrocyte. Section of the lamina terminalis taken at the point of ventriculostomy shows the ependymal lining thickened, with the ependymal cells assuming a strati-

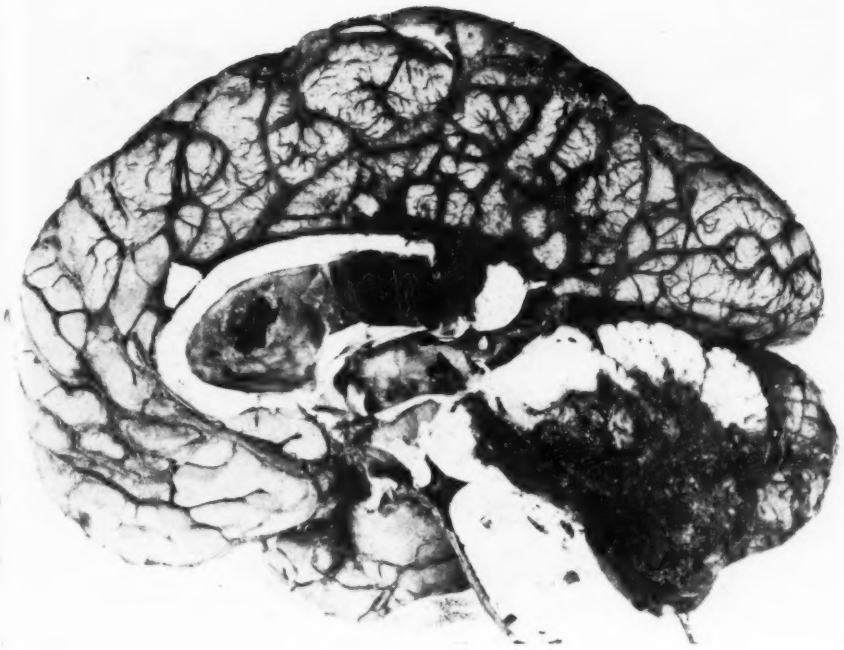

FIG. 4. Site of tumor in cerebellum.

fied appearance. All of the surface of the removed tissue is lined by this epithelium. Diagnosis: Hemangioma with reactive gliosis."

In 1931, 15 years prior to this patient's death, an unintentional ventriculostomy was done through a misinterpretation of the findings in the chiasmal region at operation. The stoma remained patent. It relieved pressure sufficiently to allow considerable return of vision. The tumor may or may not have been removable in 1931 had the correct diagnosis been made. The block to the cerebrospinal fluid circulation was complete enough (though unrecognized) at the 1st operation to cause a considerable internal hydrocephalus and ballooning downward of the 3rd ventricle into the chiasmal region. If at the time of the 1st operation the tumor could not have been removed, as much probably was accomplished in prolonging life as a surgical attack on the tumor itself. Nevertheless there came a time when, in spite of a functioning ventriculostomy stoma, the growth of the tumor upset the balance, and symptoms

of pressure returned. Neither at the 1st nor at the 2nd hospital admission were there any focal symptoms or signs of localizing value.

SUMMARY

1. A case is reported of a 3rd ventriculostomy performed through the misinterpretation of a lesion exposed at operation.

2. There was postmortem proof that the ventriculostomy stoma remained patent for the 15 years that the patient lived.

3. In spite of the functioning stoma, the growth of the cerebellar tumor brought about a return of symptoms.

4. Operations designed to by-pass ventricular obstruction will find their greater use in static obstructions rather than those caused by progressive lesions.

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THE CRANIAL LOCALIZER

ALFRED UIHLEIN, M.D., AND DANA A. ROGERS
Section on Neurologic Surgery, and Instrument Shop, Mayo Clinic, Rochester, Minnesota

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The need for an instrument that can be used as a localizer in neurosurgical operations on the brain has been recognized. Such an instrument must be of light but sturdy construction. When applied to the head, it must not jeopardize the visualization of any portion of the calvarium. The instrument must be adjustable. When it is in position it must be so firmly attached that motion of the patient's head and the surgical drapes covering the head will not disturb the position of the localizer in any way.

We feel that we have designed such an instrument. We have designated it the "cranial localizer." The instrument is simply constructed and can be adjusted readily to the head of any patient (Fig. 1). A roentgenogram from the anteroposterior or lateral view is made first to check accurately the position of the instrument with relation to definite cranial bony landmarks. This roentgenogram of the cranial localizer superimposed upon the skull (Fig. 2) aids in localization of intracranial pathologic processes prior to operation. For teaching purposes, it seems to have special

value as it assists the student to visualize the location of the cerebral hemisphere and the affected region in relation to the localizer, thereby facilitating the explanation of the rationale of various operative procedures.

The feature of this simply constructed instrument is that it is similar to any spectacle

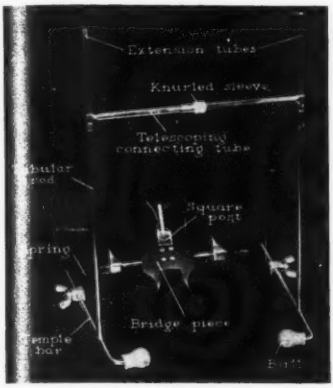

FIG. 1 (*left*). Cranial localizer. Note particularly the simple construction and means of adjusting to fit the head of any patient.

FIG. 2 (*right*). Lateral view of cranial localizer superimposed on skull of patient.

frame (Fig. 3). A square post on the bridge piece receives a round rod which is attached to the frame. This sliding joint permits vertical adjustment and it is secured by a thumbscrew. The temple bars terminate in balls which fit the external auditory canals. Thus the bridge piece over the nose and the temple bars, held in place by the balls fitted into the external canals, are fixed points when the localizer is on the head of the patient (Fig. 3). Each temple bar is joined to the frame by a spring steel arm bent to an angle of 90°. Each end of the spring has a block with hole and thumbscrew. One end of the spring fits on the frame while the other end receives the temple bar. This permits adjustment of the width of the frame as well as the length of the temple bars. A short vertical post is located on each temple bar about 1 inch forward (an arbitrary distance) from the external auditory canal. These short posts, which are perpendicular to the temple bars, receive a longer tubular rod to which extension tubes may be added if desired. The tubular rods may then in turn be joined by a telescoping connecting tube for stability and localization. A knurled friction sleeve fits on the telescoping connecting tube. This sleeve is used as an indicator for the center point when the cranial localizer is completely assembled.

FIG. 3. Cranial localizer adjusted to patient's head. The bridge piece and the balls on the ends of the temple bars are fixed points when the localizer is in position.

After the localizer has been placed on the head of the patient adjustments must be made, by using the thumbscrews, to secure the cranial localizer properly to the head. These screws adjust the width of the frame and the length of the temple bars. By sliding the frame up or down on the vertical post on the bridge piece, the temple bars can be made to lie in a posi-

tion parallel to the floor of the anterior cranial fossa. It is possible to estimate this position by having the temple bars lie parallel to the downward slope of the distal third of the eyebrows at the outer canthi of the eyes. This slope of the eyebrows almost exactly parallels the slope of the floor of the anterior cranial fossa. A roentgenogram from the lateral view, with the localizer superimposed over the skull, permits final accurate adjustment of the localizer frame to the desired position (Fig. 2). When the temple bars are parallel to the floor of the anterior fossa the upright tubular posts will be parallel with, although posterior to, the coronal suture. In this position the vertical tubular rods do not impede the surgeon and the localizing value is increased.

This instrument has been used in several types of neurosurgical cranial operative procedures, particularly in prefrontal lobotomy. Its value in this operative procedure has been discussed in a recent publication.[1] It has been used by surgeons other than the senior author. Favorable comments prompted the authors to prepare this report. It is believed that the cranial localizer can be a valuable aid to the surgeon, his assistants and students in the localization of craniotomy procedures. This is a preliminary report; some modifications already have been made and will be included in a later report.

SUMMARY

An adjustable apparatus called the "cranial localizer" has been described which, the authors feel, offers considerable assistance as a surgical guide in carrying out various cranial neurosurgical procedures. The apparatus is of simple construction, is lightweight and can be sterilized in its entirety or in parts. When placed on the head of the patient it cannot be readily dislodged even with the operative field surrounded by sterile drapes. The cranial localizer will assist the surgeon to identify quickly skeletal and intracranial landmarks that are so important in the execution of a successful neurosurgical intracranial procedure.

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A CERVICAL AND A LUMBAR PILONIDAL SINUS COMMUNICATING WITH INTRASPINAL DERMOIDS

REPORT OF 2 CASES AND REVIEW OF THE LITERATURE

ERNEST SACHS, JR., M.D.,* AND GILBERT HORRAX, M.D.

Department of Neurosurgery, The Lahey Clinic and the New England Deaconess Hospital, Boston, Massachusetts

(Received for publication May 10, 1948)

PILONIDAL sinuses, also called congenital dermal sinuses, are among the most common congenital defects of the midline in the lumbar region, but occasionally may be present at higher levels. It is not generally appreciated, however, that sinuses of this kind, instead of being harmless blind tracts, may in rare instances communicate directly with the subdural or subarachnoid space at any level of the spinal column, and still more rarely, may terminate in an intraspinal tumor. Knowledge of this fact in patients having spinal cord symptoms in the presence of a pilonidal sinus, *particularly over the higher spinal segments*, should lead to earlier surgery with less disability in a group of patients whose ultimate prognosis should be excellent if the tumor is removed before serious cord damage has taken place.

The present communication serves to put on record two examples of these uncommon intradural dermoids as well as to review the literature concerned with these tumors. Comprehensive reviews on the subject of spinal dermoids are to be found in papers by Fraser,[15] Gross,[18] Bradford,[6] Boldrey and Elvidge,[4] List[28] and Craig.[11] We wish to add our own case here, and one other case, histologically verified and previously unreported, with which we are familiar, thanks to Dr. Leonard T. Furlow. This brings the total number of spinal dermoids and epidermoids in the literature to 61 (Table 1). We have reviewed all the reported cases to which references are available, but have quoted freely from Boldrey and Elvidge, Craig, and Bradford on cases unavailable to us. The source of all cases in the tables is given in the bibliography. Rasmussen, Kernohan and Adson[37] mention 2 cases, Steinke[43] mentions 2, and Elsberg (1941) mentions 4 of his own, none of which is included here, since no details are given and it cannot be determined which of these has been previously reported.

NOMENCLATURE

The nomenclature of these tumors has passed through various phases which we shall summarize briefly for the sake of clarity. Greater detail on this score can be found in the papers of Bostroem,[5] Bailey,[3] Horrax,[22] and in the thorough studies by Critchley and Ferguson.[12]

Cruveilhier first introduced the term "tumeur perlée" in 1829 when he

* Fellow in Neurosurgery, The Lahey Clinic, Boston, Massachusetts.

reported 2 cases. In 1838, Johannes Mueller added 2 more cases and gave them the specific name "cholesteatomas" because of the cholesterin crystals he found. In 1855 Virchow, in his studies "Ueber Perlgeschwülste" advocated a return to Cruveilhier's terminology because all such tumors did not contain cholesterin. Bostroem[5] introduced the name "pial dermoids and epidermoids, and dural dermoids," based on thorough anatomical studies of the genesis of these tumors. This classic paper did not gain full recognition, as far as the use of these terms was concerned, for many years. Bailey[3] advocated a return to Cruveilhier's "tumeur perlée," as it has the advantage of being descriptive and non-committal. He also did not accept Bostroem's work as showing conclusively that they were epidermal in nature or pial in origin. In 1922, one of us (G. H.)[22] suggested that Bostroem's names were too cumbersome, and that the use of the collective descriptive name "cholesteatoma" would be preferable. By designating them as "meningeal," no confusion would exist with those tumors arising from rests in the pituitary region, or with those composed of detritus arising from chronic middle ear disease. By the expedient of calling them "hair-containing" and "non-hair-containing," the dermoids and epidermoids could be differentiated.

Harvey and Burr[20] showed that the views of His and Kölliker, who maintained that the meninges had a common derivation from the mesoderm, were incorrect. The former authors proved that certain ectodermal elements derived from the neural crest helped to form the mesenchyme, and took part in the formation of the pia and arachnoid. This immediately made it clear that dermoid tumors could arise as "inclusions" in ectodermal tissue. Ingraham and Bailey[24] used the terms "teratoma" and "teratoid" to describe all these tumors, and did not differentiate the dermoids from the teratoids. Hosoi[23] felt that the 10 spinal "teratomas" he reviewed were bidermal only, so he called them teratoid. Since then, Bucy and Buchanan[7] essentially agreed with Hosoi, but Masten[32] reported a case which she felt was truly tridermal in origin, and quoted a case of Voss (1934), as well as the case of Puusepp (1938), which also seem authentically tridermal. In many of these complex tumors, derivatives of one or two germ layers tend to overgrow the others and final analysis of the total number of germ layers present may be exceedingly difficult. The endodermal derivatives seem to be most difficult to identify, and are subject to considerable controversy.

In this paper we shall use the following classification.

Epidermoid: Tumors having only epidermal tissue and its debris, and definitely *lacking* the dermal structures. They may be traumatic or embryonic in origin, and produce characteristic growths such as the so-called cholesteatomas or pearly tumors.

Simple dermoid: Tumors consisting of epidermis, dermis and dermal glands. They may also have a pearly sheen, but as a rule not so striking as that of the epidermoids.

Teratoid: Tumors with recognizable tissues of two germ layers.

Teratoma: Tumors with recognizable tissues and complex organs derived from all three germ layers.

DERMOIDS AND EPIDERMOIDS (Table 1)

Verification of all the reported dermoid and epidermoid cases is difficult and some question of the authenticity of 6 of them has been cast by Craig,[11] who did not feel that the cases of Eppinger (1875), Ivanow (1903), Raymond *et al.*,[38] Frick (1911), Elsberg,[13] and 1 of those of Naffziger and Jones[34] were true dermoids or epidermoids. The case of Frick (1911) was originally reported as a teratoma and is included here under that heading, although Boldrey and Elvidge[4] classified it as a dermoid in their review in 1939. The 4th case of Naffziger and Jones[34] contained elements of two germ layers and hence is listed here as a teratoid. The case of Walker and Moore,[44] which was reported as a "teratoid tumor," is included here as a "dermoid," as Craig[11] and Masten[32] have done. We are including as "dermoids" 2 cases reported by Ingraham and Bailey,[24] and 3 cases reported by Bailey and Ingraham[2] which they classified as teratoids. These tumors had elements of one germ layer only.

Mahoney[30] reported 142 dermoids and epidermoids of the entire central nervous system, and Rand and Reeves[36] estimated that not more than 200 dermoids had been reported in the literature. Therefore, the spinal dermoids and epidermoids probably constitute about one-fourth of the total number of those found in the central nervous system.

PILONIDAL SINUSES COMMUNICATING WITH THE DURA

Simple pilonidal sinuses in the lumbar region are seen frequently, but those communicating with the dura are even rarer than the above-mentioned dermoids and epidermoids. Walker and Bucy[45] in 1934 found only 7 such cases in the literature, and in 1942 Kooistra[26] found 5 more and added 1 of his own, bringing the total to 13. Since then the cases reported by Ingraham and Bailey,[24] Bailey and Ingraham,[2] Cliffton and Rydell,[9] and the case of Walker and Moore[44] not included in Kooistra's review, together with the 2 cases reported here make a total of 22 cases that we have been able to find in the literature to date. Of the 22, only 14 were connected with an intraspinal tumor (Table 2).

Kooistra pointed out that only 13 pilonidal sinuses had been reported above the sacrococcygeal region, and of those communicating with the dura there were but 9 above the lumbar region. Only 1 of the latter was cervical (Clark, 1918), and this case was never confirmed by operation or by autopsy.

TERATOMAS AND TERATOIDS

We are also reviewing all the reported spinal teratomas (Table 3) since clinically these cases are very similar to the above. Masten[32] found 13 in the literature and added 1 case, making 14. French and Peyton[16] brought

TABLE 1. *Intraspinal dermoids and epidermoids*

Author	Year	Sex	Age	Type	Location	Operative Result	Remarks
1. *Eppinger	1875	?	?	Chol.	Mult. cord & brain	?	Not accepted by Craig[11]
2. Chiari	1883	M	33	Chol.	T4–6	—	Intramed. Called "dermoid" by Bostroem
3. *Muscatello	1893	F	5 mos.	Der. & chol.	L5	—	Bifid thoracic spine
4. *Trachtenberg	1898	M	55	Epi. & der.	C-Th-L	—	Mult. cholest. brain also
5. White & Fripp	1900	M	30	Der.	T2-4	Died—2nd op.	Also had Hodgkin's
6. *Ivanow	1903	?	Birth	Der.	All cord	—	Anencephalic monster. Intramed. tumor
7. Raymond	1904	M	32	Der.	Mult.	—	Mult. fatty lesions cord. Not accepted by Craig[11]
8. *Berkal	1906	F	27	Chol.	Conus	—	Intramed.
9. *Harriehausen	1909	F	23	Der.	L1	—	Double cord below tumor
10. *Verebely	1913	M	15	Der.	L?	—	
11. Robertson & Ingham	1916	F	19	Chol.	L?	—	
12. Elsberg	1916	F	60	Der.	C4 to brain	Improved	Microphoto. "gliomatous? cyst." Not accepted by Craig[11]
13. *Guizetti	1921	F	30	Der.	L5-S1	Improved	
14. *Lauterberg	1922	M	7	Epi.	Cauda	—	Incidental autopsy finding
15. *Marinesco & Draganesco	1924	M	30	Chol.	T10-11	"Improved"	Recurred 7 yrs. Postop. death
16. *Dal Bo	1926	F	42	Der.	T7-S	—	17 cm. in length
17. *Melnikoff-Raswednenkoff	1926	F	26	Epi.	T5-6	—	Intramed.
18. *Salotti	1927	M	31	Der.	T12	Died	Septicemia from decubitus ulcers
19. Shallow	1928	M	30	Der.	L?	Improved	
20. Sharpe & Sharpe	1928	M	3	Der.	T11	Improved	Communicating infected sinus
21. Critchley & Ferguson	1928	M	15	Epi.	T7-8	Improved	
22. *Delrez	1929	F	5	Der.	L3-S	Improved	Also sarcoma kidney
23. Fraser	1930	F	22	Epi.	T11	Improved	
24. *Pitotti	1930	M	26	Der.	S	—	
25. Craig & Mitchell	1931	M	8	Epi.	T12-L1	Improved	
26. *Michelson	1932	F	32	Chol.	T11-12	Died	Recurred 3½ yrs. Postop. death
27. *Schroeder	1932	F	35	Chol.	T10-11	Improved	
28. Mixter	1932	M	23	Der.	C2-5	Improved	Intramed. recurrence
29. ‡Bouchut *et al.*	1932	M	71	Epi.	Conus	—	Intramed. tumor, disembryoplasia cord, & syringomyelia
30. Hipsley	1933	F	3	Der.	T1-2	Improved	Communicating sinus
31. †Ottonello	1933	F	20	Der.	T2-4	?Improved	Rachischisis; also sinus
32. Gross	1934	M	44	Epi.	T2-3	"Improved"	Intramed. tumor; recurred 3 yrs.
33. Naffziger & Jones	1935	F	60	Der.	T12-S2	Improved	Incomplete removal but good recovery
34. Naffziger & Jones	1935	F	27	Chol.	T12-L1	Improved	
35. Naffziger & Jones	1935	F	35	Chol.	L3-4	Improved	
36. Puech *et al.*	1936	M	36	Chol.	T6-7	Improved	Intramed.
37. Love & Kernohan	1936	M	40	Der.	L-S	No improvement	Intramed.
38. *Varshaver	1937	?	?	Chol.	?	?	
39. Stammers	1938	M	2	Der.	L5	Improved	Communicating sinus
40. Bradford	1938	M	3	Der.	L2-5	Improved	Intramed.
41. Masson	1938	M	38	Der.	T8	Improved?	Pain relieved but paresis worse
42. Boldrey & Elvidge	1939	M	17	Epi.	L2-4	Improved	
43. Boldrey & Elvidge	1939	M	45	Der.	L3-4	No change	"Anal fistula" thought pilonidal sinu
44. Boldrey & Elvidge	1939	M	2½	Der.	T12-S2	Improved	Communicating sinus & recurring attacks meningitis

TABLE 1—*continued*

	Author	Year	Sex	Age	Type	Location	Operative Result	Remarks
45	‡Verbeist	1939	M	65	Epi.	T12	No improvement	Postop. sterile meningitis
46	King	1939	M	17	Epi.	L4-5	No improvement	Had total of 6 ops.
47	Walker & Moore	1939	M	3	Der.	T9-10	Died	Called teratoid but ecto. derivative. Dimple & sinus
48	Bagley & Arnold	1940	M	9	Der.	T12-S2	Improved	
49	List	1941	M	14	Epi.	L3	Improved	
50	List	1941	M	47	Epi.	L3-4	Improved	
51	List	1941	M	8	Der.	T12-L2	Improved	
52	Kooistra	1942	M	19	Der.	C7-T4	Improved	Communicating sinus. Kooistra calls intramed. cyst with sebaceous matter & hair
53	French & Peyton	1942	M	2	Epi.	T12-S	Improved	Communicating sinus
54	Craig	1943	M	25	Epi.	T11	No improvement	
55	Ingraham & Bailey	1946	M	Birth	Der.	C-Th	Improved	Wrinkled skin & spina bifida over tumor
56	Ingraham & Bailey	1946	M	6½	Der.	Th	Improved	Dimple with sinus tract into canal
57	Bailey & Ingraham	1947	F	10	Der.	T7-L4	Improved	Histolog. verified by cyst fluid only
58	Bailey & Ingraham	1947	F	3½	Der.	Th-L	Improved	Histolog. verified by cyst fluid only
59	Bailey & Ingraham	1947	M	?	Der.	Conus	Improved	Histolog. verified by cyst fluid only. Sinus tract to tumor
60	Sachs & Horrax	1949	M	35	Der.	C2-7	Improved	Communicating sinus
61	Furlow (reported by Sachs & Horrax)	1949	M	3	Der.	L4-5	Improved	Communicating sinus from dimple

* Quoted from Boldrey and Elvidge.[4]
† Quoted from Bradford.[6]
‡ Quoted from Craig.[11]

the number to 16. Ingraham and Bailey,[24] in a paper on teratoids and teratomas in infancy and childhood, reported 7 intraspinal tumors: 2 dermoids, 2 teratoids and 3 teratomas. In 1947, Bailey and Ingraham[2] added 6 more intraspinal cases: 3 dermoids and 3 teratoids. The "dermoids" were cystic tumors histologically identified by epithelial cells in cyst fluid only. Since we are including the 4th case of Naffziger and Jones[34] as a teratoid, this brings the total number to 25.

We do not propose to deal with the etiology and genesis of these various tumors other than to say that there is a considerable body of evidence to show that the epidermoid and dermoid tumors arise from faulty closure of the neural crest with inclusion of ectodermal elements, while the teratoid and teratomatous tumors arise from maldevelopment of the ovum or the inclusion in early embryonic life of multipotential cells (teratoids), or totipotential cells (teratomas).[7] For further discussion of this side of the subject reference should be made especially to the articles by Bostroem,[5] Craig,[11] Bradford,[6] Fraser,[15] and Ingraham and Bailey.[24]

TABLE 2. *Intraspinal tumors associated with dermal sinuses*

Author	Year	Sex	Age	Type	Location	Operative Result	Remarks
1. Sharpe & Sharpe	1928	M	3	Dermoid	T11	Improved	
2. Hipsley	1933	F	3	Dermoid	T1-T2	Improved	
3. *Ottonello	1933	F	20	Dermoid	T2-4	?	
4. Stammers	1938	M	2	Dermoid	L-5	Improved	
5. Boldrey & Elvidge	1939	M	45	Dermoid	L3-4	No change	Believed anal fistula & sinus
6. Boldrey & Elvidge	1939	M	2½	Dermoid	T12-S2	Improved	
7. Walker & Moore	1939	M	3	Dermoid	T9-10	Died	Called teratoid, but only 1 germ layer
8. French & Peyton	1942	M	2	Epi.	T12-S	Improved	
9. Kooistra	1942	M	19	Dermoid	C7-T4	Improved	Called "intramed. cyst with sebaceous matter & hair"
10. Ingraham & Bailey	1946	M	6½	Dermoid	Th	Improved	Dimple with sinus tract into canal
11. Bailey & Ingraham	1947	M	?	Dermoid	Conus	Improved	
12. Bailey & Ingraham	1947	F	1/12	Teratoid	S	Improved	
13. Sachs & Horrax	1949	M	35	Dermoid	C2-7	Improved	
14. Furlow (reported by Sachs & Horrax)	1949	M	3	Dermoid	L4-5	Improved	

* Quoted by Bradford.[5]

Summary

Males = 11, females = 3
Average age = 9 years
Cervical = 1, thoracic = 7, lumbar = 4, sacral = 2
Improved = 9, no change = 1, death = 1, unstated = 3

TABLE 3. *Intraspinal teratoids and teratomas*

Author	Year	Sex	Age	Type	Location	Operative Result
1. *Gowers	1876	?	Adult	Teratoid	Conus	—
2. *Gerlach	1894	M	36	Teratoid	C	—
3. *Forbes	1905	M	5½	Teratoid	C	—
4. *Frick	1911	F	41	Teratoid	L2-5	—
5. *Andre-Thomas & Querey	1912	—	—	Teratoid	C & Th	—
6. *Bielschowsky & Unger	1920	M	37	Teratoid	C3	—
7. Hansmann	1926	F	21 days	Teratoid	S	—
8. Kubie & Fulton	1928	M	2	Teratoma	T9-L1	Improved
9. Kubie & Fulton	1928	F	27	Teratoma	C3-C4	Improved
10. Hosoi	1931	M	24	Teratoid	L2-L3	Improved
11. †Voss	1934	?	?	Teratoma	?	?
12. Bucy & Buchanan	1935	M	2¾	Teratoid	T11-L4	Improved
13. Naffziger & Jones	1935	M	45	Teratoid	L1-S	Improved
14. †Puusepp	1938	?	?	Teratoma	?	?
15. Masten	1940	F	5	Teratoma	C4-T1	Died
16. French & Peyton	1942	M	36	Teratoma	L3-L5	Improved Recurred
17. French & Peyton	1942	M	7	Teratoma	T8-S	Improved
18. Ingraham & Bailey	1946	F	4 mos.	Teratoid	C	Improved
19. Ingraham & Bailey	1946	F	2½	Teratoma	C2-Th	Improved
20. Ingraham & Bailey	1946	M	5	Teratoma	L	Improved
21. Ingraham & Bailey	1946	F	1	Teratoma	L	Improved
22. Ingraham & Bailey	1946	M	2	Teratoid	L	Improved
23. Bailey & Ingraham	1947	F	Birth	Teratoid	Cauda	Improved
24. Bailey & Ingraham	1947	F	1/12	Teratoid	S	Improved
25. Bailey & Ingraham	1947	M	3	Teratoid	L	Improved

* Quoted from Hosoi.[22]
† Quoted from Masten.[32]

CASE REPORTS

Case 1. L. G., a 35-year-old male referred by Dr. A. J. Panettieri of Bridgeport, Connecticut, was first seen at the Lahey Clinic on Feb. 20, 1947. His chief complaint was weakness and numbness of the left leg of $3\frac{1}{2}$ years' duration. His past history brought out that the patient did not walk until the age of 4. An "infection" had been noticed over the midline in the upper dorsal region at the age of $1\frac{1}{2}$, and had drained

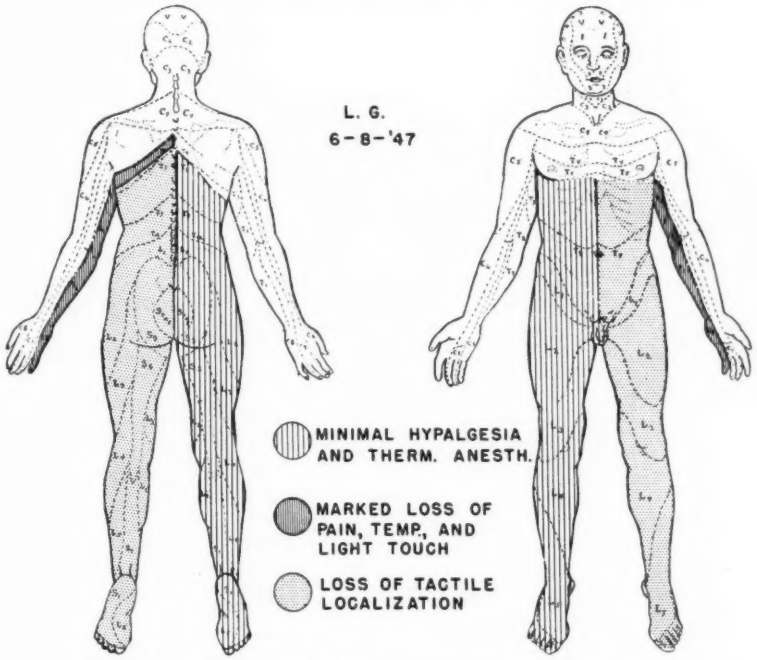

FIG. 1. Case 1. Chart showing area of sensory loss previous to operation.

periodically since that time through a midline sinus, as well as through another opening several inches to the right of this.

His present illness began $3\frac{1}{2}$ years previously with mild aching in the left knee and ankle following heavy work. Two months after this his left leg collapsed. At this time he had some numbness of the sole of the left foot and of the left little finger. He was given a medical discharge, from the service, and continued to have a mild degree of stiffness in the left hip, knee and ankle, not sufficient to keep him from light work.

Examination. Positive neurological findings: He listed to the right on forward bending. There was weakness of all muscle groups in the left leg and foot. Gait was slightly spastic but he was able to lift the left foot well off the floor in walking. Biceps and triceps reflexes were hyperactive, somewhat more so on the right. There was a positive Hoffmann on the right. Abdominal and cremasteric reflexes were absent. There was slight hypesthesia to light touch over the ulnar aspect of the left arm and

hand, and slight hypalgesia over the right side of the body up to the 4th thoracic dermatome (Fig. 1). Reflexes at ankles and knees were hyperactive. Babinski's sign was positive bilaterally. Position and vibration sense were reduced throughout the legs. There was no bladder difficulty. There was an old, healed scar over the lower cervical vertebrae in the midline, and another one 2 inches to the right of this.

Lumbar puncture disclosed a complete subarachnoid block; total protein 120 mg. per cent; cells and colloidal gold curve normal. X-rays of spine showed an anomaly of the lower cervical region with almost complete fusion of the bodies of the 6th and 7th vertebrae and narrowing of the intervertebral space between the 7th cervical and the 1st thoracic vertebrae.

FIG. 2. Case 1. Gross specimen of sinus tract leading from the skin down to the intraspinal dermoid.

The patient was admitted to the New England Deaconess Hospital on June 8, 1947. There was no change in the findings since the first examination, but he was becoming more constipated than ever before. A pantopaque myelogram revealed a complete block, the lower end of which was between the 2nd and 3rd thoracic vertebrae.

Operation (G. H.), June 12, 1947. The spines and laminae of the 5th, 6th and 7th cervical and 1st and 2nd thoracic vertebrae were removed. The sinus tract in the skin at the 1st thoracic level was picked up and dissected out as the operation proceeded (Fig. 2). It extended from the skin diagonally upward and passed through the spinous process of the 7th cervical vertebra. After the spines and laminae had been removed, the sinus tract could be followed down to the dura at about the level between the 6th and 7th cervical vertebrae.

The dura felt firm to palpation and there was no pulsation visible. Upon opening the dura a somewhat elliptical-shaped tumor was at once apparent occupying the left side of the spinal canal, compressing the cord into almost ribbon-like thinness downward and to the right (Fig. 3). The tumor was situated beneath the arachnoid, and was yellowish-white in appearance with somewhat of a pearly sheen, although not as lustrous as epidermoids. In attempting to lift the growth up at its lower end and remove it intact, its delicate friable containing membrane broke immediately, so that it had to be removed piecemeal. The tumor consisted of material resembling, but somewhat firmer than, cream cheese and contained numerous fine, long light brown hairs (Fig. 4). It extended upward beyond the bony opening for a distance of

about 2½ inches, i.e., to about the level of the atlas. All the contents of tumor above the bony opening were carefully spooned or sucked out.

The dura was reclosed completely, as were muscles, fascia and skin in layers with silk. Penicillin was instilled intra- and extradurally.

Postoperative course was smooth and patient was up on the 12th day with some residual weakness of the arms, the left being weaker than the right. At no time did he have any urinary difficulty nor any evidence of infection in the subarachnoid space. He was discharged on 19th postoperative day, still gaining strength in arms and legs.

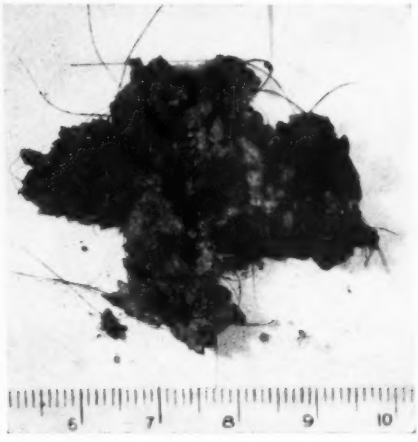

FIG. 4. Case 1. Cream cheese-like contents of tumor containing numerous long, fine hairs.

FIG. 3. Case 1. Photograph of the tumor at operation: it extended upward (cephalad) somewhat beyond the dural opening. The sinus tract has been grasped by forceps and can be seen passing into the upper end of the exposed tumor. The spinal cord has been compressed over against the right-hand wall of the spinal canal and is not visible.

On Dec. 11, 1947, 6 months postoperative, the patient reported for check-up examination. He was able to walk by himself 6 weeks after operation and said that his legs were now about as strong as they ever were. He had still some numbness of left hand and arm and the left hand grip was rather weak. The right hand and arm were of normal strength. His only sensory change was hypalgesia of left hand and arm up to the elbow (Fig. 5). He had been back at his regular work as an assembler for the past 2 months.

Pathologic Report. The gross specimen consisted of a mass of friable yellowish material mixed with abundant curly light brown hairs, together with some mem-

branous material also bearing fine hairs and measuring 8×0.6×0.2 cm. Included as well were the skin and sinus tract (Figs. 2 and 4).

Microscopic Description. The cyst wall showed a lining of stratified squamous epithelium beneath which were sebaceous glands and hair follicles scattered in a dense collagen background (Figs. 6 and 7). In one area there were many hair follicles embedded in dense collagen. In other sections there was necrotic acellular material resembling that seen in epidermal inclusion cysts.

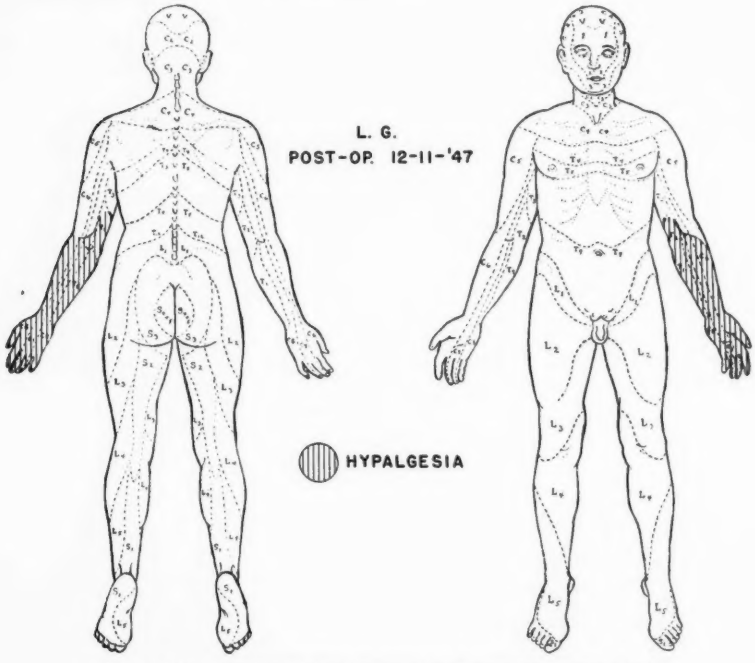

FIG. 5. Chart showing minimal sensory loss 6 months postoperatively.

Through the courtesy of Dr. Leonard T. Furlow, we are reporting another case of pilonidal sinus associated with intraspinal dermoid, on which he operated in 1935.

Case 2. A $2\frac{1}{2}$ -year-old male child was referred to Dr. Leonard T. Furlow of St. Louis, Missouri on Sept. 18, 1935.

History. He was known to have had a small, dimpled area in the median line of the back in the lower sacral region since birth. About $3\frac{1}{2}$ weeks before being seen by Dr. Furlow, he had fallen, striking the lower part of his back, and complained of pain in the region of the dimple. The day after the fall some swelling and tenderness had been noted around the dimple, and within the next 2 or 3 days he complained of headache, pain in the back and in his legs. He was admitted to the Children's Hospital of St. Louis with a fever of 38.5 C.

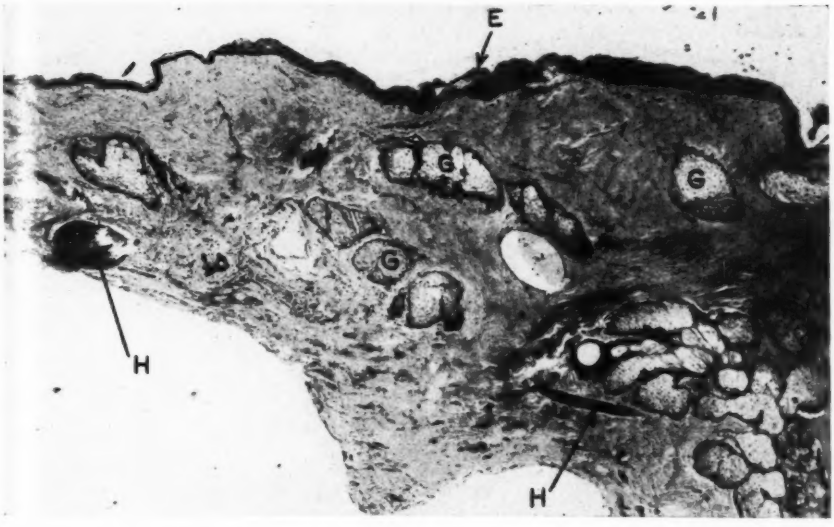

FIG. 6. Case 1. Capsule of dermoid lined by stratified squamous epithelium (E), with sebaceous glands below this lining (G), as well as hair follicles (H). (×40)

Examination. There was moderate stiffness of the neck and spasticity of the spinal muscles. Severe pain was present on moving the legs, particularly when the thighs were flexed on the abdomen. In the midline of the back over the lower sacral

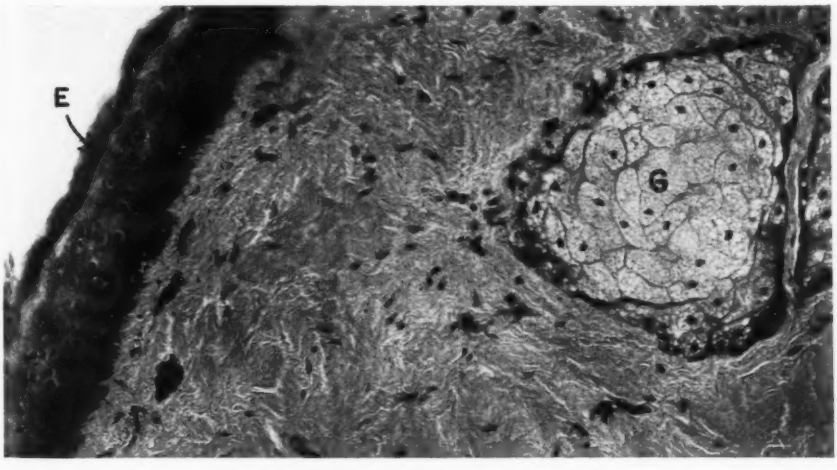

FIG. 7. Case 1. High power photomicrograph of cyst capsule to show more detail of stratified epithelium (E) and sebaceous glands (G). (×200)

region there was a hard, tender mass with a suggestion of fluctuation on deep palpation.

Lumbar puncture on admission revealed slightly turbid fluid containing 325 cells per c.mm. The next day the spinal cell count had risen to 1900, the majority being polymorphonuclear leukocytes. No organisms were present by smear or cul-

FIG. 8. Case 2. Photomicrograph of cyst showing (A) amorphous material within cyst wall; (E) epithelium with its four strata; and (c) dermal layer (corium). (×165)

ture. The child was treated by antimeningococcus serum intramuscularly and intraspinally without benefit.

On Sept. 18, 1935, Dr. Furlow inserted a large needle to one side of the inflamed mass in the sacral region and evacuated a few flecks of cloudy material followed by cloudy cerebrospinal fluid. This fluid showed many pus cells but no organisms. It was felt that the lesion might be a dermoid extending into the subarachnoid space and exploration was advised.

Operation (Dr. Furlow). On Sept. 19, 1935, the subcutaneous portion of the cystic mass was removed together with an area of overlying skin. It was found that the lower end of this sac merged into a thin stalk, and this stalk could be seen extending down in the median line between the 4th and 5th lumbar spines. A laminectomy was

performed, and when the dura was exposed it was seen that the stalk passed through the dura into the subarachnoid space. The stalk and a small area of dura contiguous to it were excised, leaving a small opening through which cholesteatomatous material was extruded. The dura was opened sufficiently so that all of this material could be removed, after which the dura was closed completely, as were the muscles and other tissues.

Course. The child made an uneventful recovery and was discharged apparently very well on Oct. 2, 1935.

On April 19, 1936, 6 months after operation, he was apparently perfectly well with no sensory or motor changes and normal sphincter control.

Comment. It was Dr. Furlow's opinion that the child had a small dermoid in the lumbosacral area within the dura which connected by a stalk and sinus through the dura to the skin, and that as a result of the fall some of the contents of the dermoid were forced through the stalk into the subarachnoid space, setting up a chemical irritation giving meningeal symptoms.

Pathologic Report. Two specimens of tissue were submitted for microscopic study. The first was a cross section of the sinus tract with a central core of epithelium within which there was a space containing amorphous material.

The second represented the capsule and contents of the cyst. This showed a layer resembling normal skin cut longitudinally with epithelium having the stratum corneum, lucidum, granulosum and germinativum. On one side of this epithelium there was amorphous as well as hyalinized material with areas of apparently empty cell spaces. On the other side of the epithelium there was a thick layer of derma (corium) in which there were a few capillaries (Fig. 8).

The microscopic diagnosis was a dermoid even in the absence of hair, inasmuch as both dermal and epidermal layers were present.

DISCUSSION

There are a number of unusual features concerned with Case 1. This patient apparently represents the first reported instance of a verified cervical pilonidal sinus communicating with an intraspinal dermoid. It is also interesting to note that this man had had a draining sinus over the upper part of his back since the age of $1\frac{1}{2}$ years, had not walked until he was 4 years of age and yet never had serious disability at any time, even when he came in for treatment. It seems evident that he was having symptoms of his subarachnoid lesion for 31 years, but had adjusted to the profound degree of spinal cord compression due to the extremely slow increase in size of the tumor. A further point of interest is the fact that this patient was 35 years of age when he came for treatment. This is far above the usual age of patients with congenital dermal sinuses communicating with intraspinal tumors. As will be seen in Table 2, the average age for these patients is 9+ years. Of the previously reported patients, 8 were under the age of 4.

Of the total 14 cases of pilonidal sinuses communicating with intraspinal tumors, it is interesting that the sex incidence has been 11 to 3 in favor of males. Only 1 patient died, all of the others having improved to some degree after surgery, with the exception of Boldrey and Elvidge's first case, and the case of Ottonello in which the end result was not given. In 12 of the total 14

cases with communicating sinuses the intraspinal tumor was a dermoid. Bailey and Ingraham's case[2] is the only one ever reported that was associated with a teratoid.

Only 2 dermoids of the cervical region of the cord (Elsberg, 1916; Mixter, 1932), and only 1 other pilonidal sinus of the cervical region (Clark, 1918) have been reported. The case of Clark was never substantiated by operation or autopsy. Eight of the 25 teratomas were cervical, a higher percentage of cases than of the other types of the tumors under discussion.

The chance association of a pilonidal sinus with an intraspinal dermoid, though rare in this combination, is typical of the tendency of midline anomalies of the spinal cord, dura, vertebral spines, laminae, vertebral bodies, and the overlying soft tissue and skin to occur in various combinations.

Obstruction of a discharging pilonidal sinus in communication with the dura may account for sudden symptoms of myelitis either from direct infection or chemical irritation (Craig;[11] Walker and Bucy[45]).

Draining dermal sinus tracts in the presence of neurologic symptoms in the cord should not be squeezed or injected to determine their extent, since infection may be spread intradurally.

A study of the accompanying tables reveals some hitherto unappreciated facts. Males are afflicted by these conditions in the proportion of almost 2 to 1. Dermoids occur with about the same frequency as teratomas and teratoids combined, and are about one-third more frequent than either epidermoids or sinuses with dural connection.

Dermoids are more frequent in the thoracic region. The epidermoids are about equally common in the thoracic and lumbar regions.

SUMMARY

The association of congenital dermal sinuses (pilonidal sinuses) and intraspinal dermoid tumors in 14 collected cases attests to the congenital origin of both lesions, probably due to a simple mechanical defect.

Congenital dermal sinuses in the presence of neurologic symptoms pointing to the spinal cord probably have associated intraspinal tumors.

Dermoid tumors of the cord represent approximately one-fourth of all dermoids in the central nervous system, are usually operable, and have a good prognosis.

To the 59 dermoids and epidermoids of the spinal cord previously reported we have added 2 cases, making a total of 61. There have been 25 teratomatous tumors of the spinal cord reported. In the whole group there were 22 instances in which the dural or intradural tumor was associated with a pilonidal (congenital dermal) sinus.

Case 1 presented here is the first instance of a congenital dermal sinus of the spinal cord in the cervical region communicating with the dura and with an intraspinal dermoid.

The rarity of these tumors as well as some of their striking features have been pointed out.

A brief review of the literature of intraspinal dermoids, epidermoids, teratomas and congenital dermal sinuses is made.

The authors wish to express their appreciation to Dr. Leonard T. Furlow for his permission to include his case in this report. We are also grateful to Drs. Meissner and Feldman of the pathological department of the New England Deaconess Hospital and to Dr. Louise Eisenhardt of New Haven, Connecticut, for their cooperation with the microscopic descriptions of the material.

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A TECHNIQUE FOR TRANSPLANT OF THE MUSCULOSPIRAL NERVE IN OPEN REDUCTION OF FRACTURES OF THE MID-SHAFT OF THE HUMERUS

MAX T. SCHNITKER, M.D.*
Toledo, Ohio

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FRACTURES of the mid-shaft of the humerus have associated injury to the musculospiral nerve frequently because of the peculiar course of the nerve along the bony groove approximately 7 inches long. Technical difficulties arise in dealing with the nerve when open reduction and internal fixation of the fractured humerus are desirable. This can be solved by transplanting the nerve.

The incidence of injury to the musculospiral nerve in fractures of the mid-shaft is given as about 4 per cent in civilian practice and about 30 per cent in war wounds.[3] In cases of simple transverse fractures with contusion of the nerve, spontaneous recovery occurs in most instances under proper treatment, especially with the use of the hanging cast. However, in spiral type fractures that occur in the line of the radial groove, and in comminuted fractures, complete immobilization is more difficult so that repeated trauma accentuates the original nerve damage. Ununited fractures occur most frequently in the mid-shaft of the humerus, also with an associated higher incidence of musculospiral nerve injury. Of 226 fractures of the humerus reported by Campbell,[1] 52 (23 per cent) were ununited, with an associated nerve injury in 8 cases (15 per cent).

Operative treatment is usually reserved for cases of non- or mal-union of the bone or for progressing paralysis or delayed recovery of the musculospiral nerve function. In cases requiring bony fixation, but without nerve injury, this is best accomplished through the incision of Henry[2] by splitting the brachialis muscle just lateral to the edge of the biceps and leaving the nerve buried in the posterior muscle. In the cases requiring nerve exploration or repair in conjunction with the fixation of the fracture, it is felt that the nerve should be transplanted to the medial side of the arm particularly in cases of neurorrhaphy so that the nerve will follow a shorter course under less tension, be protected by a muscular bed and be far removed from the foreign body used for internal fixation. Such a transplant can be done easily whether the nerve has been divided or even when the nerve continuity is still intact.

TECHNIQUE

The operation to be described can be done under local or general anesthesia. The entire upper arm is prepared by sterile technique and the arm abducted at the shoulder with the elbow flexed. A straight incision is made along the lateral aspect

* 1031 Secor Hotel Bldg., Toledo 4, Ohio.

of the arm from a point just proximal and posterior to the insertion of the deltoid muscle and carried distally along the external intermuscular septum toward the external epicondyle of the humerus to the groove between the brachioradialis and the brachialis muscles. (This incision is posterior to the usual one that separates the brachialis fibers.) The fascia of the lateral intermuscular septum is divided from bone distally to the origin of the brachioradialis muscle. The brachialis muscle is retracted anteriorly and the lateral head of the triceps retracted posteriorly. This will expose the musculospiral nerve lying in its bony groove. The nerve is then freed

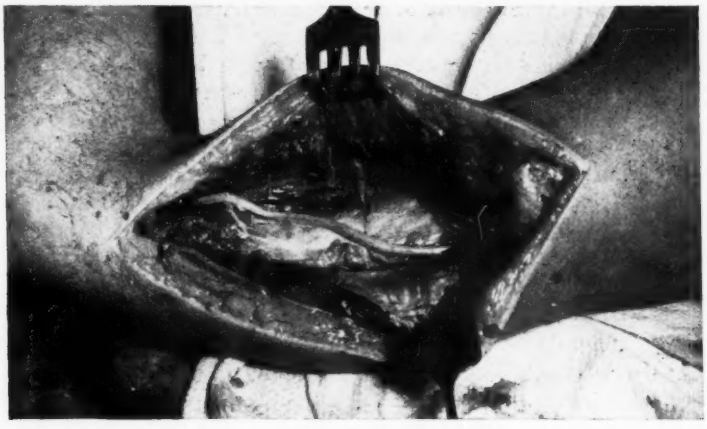

FIG. 1. Showing normal course of musculospiral nerve in its groove and overlying a hypothetical fracture. Anatomical specimen, lateral aspect left upper arm.

for its entire length, proximally to the composite teres-latissimus tendon and distally into the brachialis-brachioradialis groove, care being exercised in preserving the anterior radial recurrent artery at this point. A $\frac{1}{2}''$ linen umbilical tape is passed around the nerve and it is retracted laterally out of the way. The lateral and medial bellies of the triceps muscle are gently stripped off the posterior surface of the humerus (proximal segment) to above the upper end of the radial groove, care being taken to preserve the superior profunda artery which lies lateral to the nerve. The branch of the nerve going to the lateral head of the triceps will be seen at this point and its fasciculus can be dissected from the main nerve for several cm. if necessary to procure mobilization of the main trunk. The superficial cutaneous branch of the nerve is given off in this vicinity also, but since it runs an independent course in the loose fascia of the main trunk (Sunderland[5]), it, too, can be dissected free to the brachio-axillary angle to allow for mobilization of the main trunk, or it can be sacrificed with minimal sensory impairment. Similarly, the brachialis muscle is then gently stripped off the anterior surface of the humerus (distal segment) to the point where the branch of the musculospiral is given off to the brachioradialis muscle (Fig. 1). By gentle manipulation, the bone ends are then separated and the intact nerve is passed posterior to the upper fragment and anterior to the lower fragment with the nerve passing between the bone ends as one would pass a key chain through a key ring (Fig. 2). This places the nerve along the medial surface of the humeral shaft (Fig. 3).

FIG. 2. Showing method of transplant of musculospiral nerve from lateral to medial side of humerus through fracture site. Anatomical specimen, lateral aspect left upper arm.

A longitudinal counter incision is then made about 7 cm. long along the midportion of the medial aspect of the upper arm, along the bicipital groove. The neurovascular bundle comprising the median and ulnar nerves and brachial vessels are identified and retracted anteriorly. The musculospiral nerve is then placed posterior to the neurovascular bundle and comes to lie between the medial head of the triceps posteriorly and the attachment of the coracobrachialis muscle anteriorly (Fig. 4). In its new bed, the nerve lies between the medial border of the triceps and the coracobrachialis muscle in the upper portion of the arm and beneath the brachi-

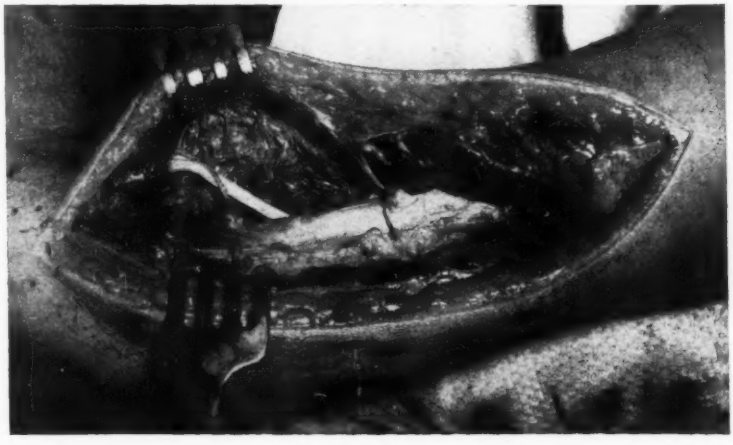

FIG. 3. Showing transplanted musculospiral nerve in its new bed along medial aspect of humerus. Anatomical specimen, lateral aspect left arm.

alis muscle to curve around its lateral border in the lower portion of the arm (Fig. 3). This transplanted position removes the nerve entirely from the lateral surface of the fracture so that fixation of the fracture with a metallic plate, bone inlay graft or on-lay grafts can be carried out without interference to later nerve function.

When using a metallic plate or on-lay graft transfixed with through-and-through screws, care must be taken that the screw point protruding through the opposite cortex is made to lie buried in muscle and not to lie adjacent to any of the nerves or blood vessels in the neurovascular bundle. Control of bleeding must be carried out meticulously, especially along the new bed for the nerve. Muscle fibers of the inner head of the triceps and the medial brachialis are then coapted over the fracture

FIG. 4. Showing tape around transplanted musculospiral nerve as it lies posterior to neurovascular bundle. Anatomical specimen, medial aspect left arm.

site and beneath the transplanted nerve to protect it from the bone fragments. Closure of both wounds is then done in the accepted manner without drainage. A well-fitting plaster cast is then applied with the hand in a cock-up position with the thumb in a grasping position and a little abducted, nearly opposite the index finger. Free movement of all fingers is imperative.

Measurements made at the operating table and on cadaver specimens show that in cases in which the musculospiral nerve has been severed, the largest gap that can be closed for an end-to-end anastomosis along the normal course of the nerve is 4–6 cm. By means of the transplant described, an additional 3–4 cm. of nerve can be gained, permitting closure of a gap up to 9–10 cm. Seddon[4] suggests sacrificing the branch of the nerve going to the brachioradialis muscle in order to gain additional nerve length, but the same gain can be accomplished by the transplant without the sacrifice of this important branch. If additional nerve length is required, the humerus can be shortened as necessary.

The question arises as to what length of nerve can be stripped safely out

of its bed without jeopardizing its blood supply. The large experience gained during the last war seemed to indicate that a large nerve could be dissected out of its bed for its entire length from joint to joint in either the upper or lower arm or leg, if gentle and meticulous technique were used. Sunderland[6] has pointed out that the intraneural vascular plexus is adequate for the entire length of a nerve providing the main nutrient arteries are preserved. In the case of the musculospiral nerve in this discussion, the most constant nutrient arteries that must and can be preserved are the profunda brachii at the upper end of the spiral groove and the anterior radial recurrent artery in the intermuscular furrow between the brachialis muscle medially and the muscles arising from the supracondylar line laterally.

SUMMARY

A technique is proposed and described for the transplant of the intact musculospiral nerve through the fissure between the bone fragments in selected cases of fracture of the mid-shaft of the humerus that require open reduction. This procedure is recommended for the complicated cases requiring bony fixation, especially if the musculospiral nerve has been injured and recovery is not complete. The transplantation of the nerve places it into a more suitable soft-tissue bed and removes it from possible further injury incident to the operative repair and healing of the fracture.

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A FOLLOW-UP REPORT OF SPINAL CORD INJURIES IN A GROUP OF WORLD WAR II PATIENTS

RICHARD C. SCHNEIDER, M.D., JOHN E. WEBSTER, M.D., AND JAMES E. LOFSTROM, M.D.

Department of Surgery, Wayne University College of Medicine, Detroit, Michigan

(Received for publication June 1, 1948)

NINETY-FOUR patients with penetrating wounds of the spine were observed and treated during the overseas period of their injuries. Two to 3 years later an inquiry was made concerning the American members of this group. The early records have been analyzed and viewed from the standpoint of the results that have rewarded efforts employed in care. Of the 94 patients, 69 were Americans and 25 were foreigners (mainly German prisoners). Follow-up reports were available in 56 of the American group.

Under pertinent headings, background data concerning the entire group of 94 cases are briefly tabulated. Several illustrations are used.

The follow-up observations are in more detail, consisting of tabulations of personal reports received in answer to letters, questionnaires, and personal examinations.

PRELIMINARY DATA

Time Interval Between Injury and Original Debridement. In 47 of the American cases there was a precise statement of the time between injury and debridement. It is of interest that 83 per cent of these patients had wound debridement within the first 24 hours after injury.

0– 6 hours	7	
7–12 hours	10	
13–18 hours	5	
19–24 hours	10	
25–36 hours	7	
37–48 hours	3	
49+ hours	5	(50, 76, 84, 96, 96)
	47	cases

Time Interval Between Debridement and Definitive Operation. In a group of 62 cases in which the time interval between debridement and definitive operation was known, it was found that 36 patients were treated sufficiently thoroughly at the time of primary debridement so that no further definitive operations were necessary. In the remaining 26 cases, the following time intervals had elapsed.

19–24 hours	2
25–48 hours	1

Within 2 weeks.....	3 (8, 9, 13 days)
Within 3 weeks.....	4 (16, 16, 18, 18 days)
Within 1 month.....	5 (23, 26, 27, 29, 34 days)
Within 3 months.....	11 (31, 31, 31, 34, 36, 37, 40, 55, 64, 76 days)
	26 cases
	36 cases (adequate operative procedures performed originally)
Total.....	62 cases

Management. A total of 38 patients of the group were treated by laminectomy at the 36th General Hospital. Two main classes of patients were encountered: (1) Those who had had no primary debridement in forward units, and (2) those on whom only a superficial wound debridement had been performed.

A uniform plan of treatment was adapted to both groups. A neurologic and roentgenologic estimation of the patient was made. A lumbar puncture with a Queckenstedt test was performed. Laminectomy was done when the roentgen study showed the presence of bone or metallic fragments within the spinal canal, when a spinal fluid block was demonstrable, or if a "dynamic" syndrome developed with progressive disability. Occasionally a spinal fluid leak through a superficially debrided open wound called for dural closure. In our experience the comminution of bone in and about the spinal canal was of a greater degree than estimated in the preoperative roentgen studies. The importance of excellent detail in the roentgenograms of the spine must be emphasized. It was noted, commonly, that the spinal wound was overlooked in the forward area, due not to a failure to obtain roentgen studies of the affected area but to distorted films, some of which failed to show even large metallic fragments in and about the canal. The rationale of debridement by laminectomy is (1) to remove metallic fragments which may, particularly when ragged, bear foreign debris, such as particles of clothing, into the spinal canal. (2) Bone fragments, which may also serve as a nidus for an inflammatory complication, may be removed. Such particles may be found within the cord substance. (3) Compression of the cord or the cauda equina by bone or a metallic fragment may be relieved. Thus surgical treatment is designed in general to prevent further damage.

Attention was focused upon the care of the patient's bladder, bowel, skin and his morale. A routine of care was provided by trained attendants on a special ward. Cystotomy drainage was provided as necessary, irrigations being done manually twice daily with boric solution. Two-hour turning was accomplished by conscientious corpsmen. Pressure areas were treated by exposure to light; enemata were used every third day if necessary. At the outset the patients were encouraged to serve their own needs as far as possible. The grouping of these patients together, under the care of sympathetic

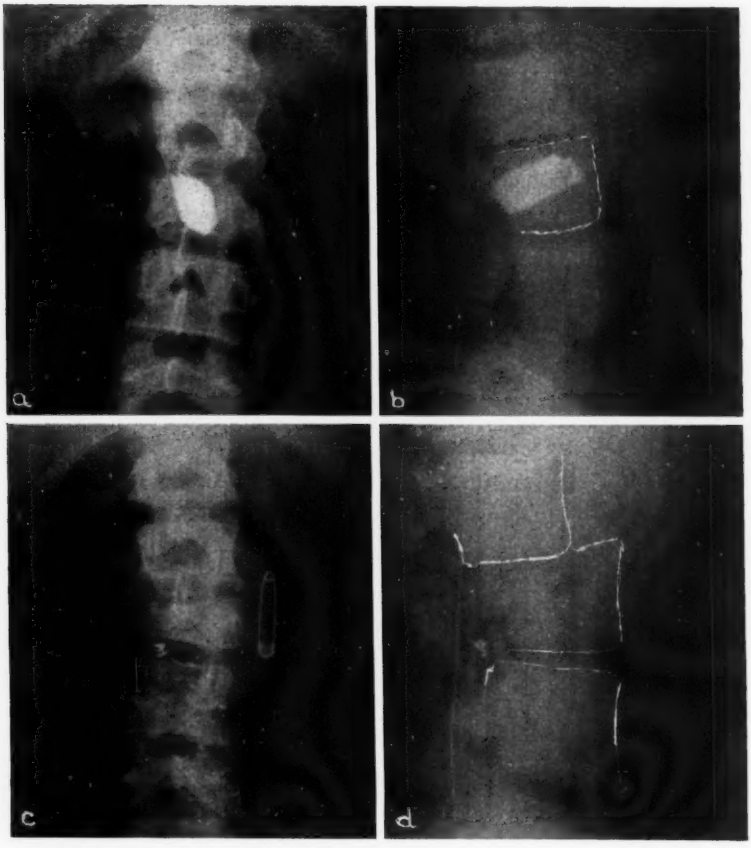

FIG. 1. (a) A large shell fragment is present in body of L3, with comminution of right half of the body and associated fractures of transverse processes of L2, L3, and L4 on the right (11–23–44). (b) Lateral view (11–23–44). (c) Postoperative removal of the large shell fragment with some loss of vertebral substance and compression of right side of the body as seen in the AP projection. (The fragment was covered with cloth from patient's overcoat.) (d) Comminution of the body is demonstrated with displacement anteriorly of a fairly large portion of right half of the body.

and understanding attendants, served, in our experience, as a major step in readjustment.

These principles in care may be applied to civilian injuries with several revisions. Urethral catheter bladder drainage with tidal irrigation should be employed in place of cystotomy drainage. Earlier catheter removal may be accomplished than could be practised in patients in the process of evacuation. Similarly, more rapid rehabilitation is now possible, although civilian facilities, even in large centers, are quite inadequate.

FIG. 2. (a) Lateral view shows comminuted fractures of bodies of L4 and L5 as a result of penetrating wound, with bone destruction occurring secondary to osteomyelitis. Catheter is outlined extending through large penetrating defect in left ileum for instillation of penicillin (1–18–45). (b) Further destruction of the body of L5 is demonstrated with considerable loss of bone detail and sequestration of anterior portion of L5 (2–24–45). (c) AP projection shows beginning productive changes of bodies of L4 and L5, and new bone formation in ligaments on the left. The large defect in left ileum is outlined. This is the result of penetrating fracture and secondary osteomyelitis (3–4–45). (d) Lateral view shows loss of the sequestrum. No further increase in destruction (3–4–45). (e and f) Complete healing of the osteomyelitis is demonstrated with solid bridging between L4 and L5 and S1. The intervertebral spaces are obliterated. The contours of the vertebral bodies are remarkably maintained in view of the extensive destruction previously seen. The defect in left ileum has decreased in size and shows no residual activity of the infection (8–20–46). (g and h) Patient 3 years after original wound. Scar of the point of entrance is seen near crest of left ileum. Range of motion is demonstrated, showing some impairment of flexion and rigidity of lower lumbar segment (3–20–48).

FOLLOW-UP DATA

In follow-up, 62 Americans were sent personal letters and questionnaires. There were 56 (90 per cent) replies. One of the patients was deceased, so that total figures on direct answers are based upon 55 cases. These follow-ups were made $2\frac{1}{2}$ to $3\frac{1}{2}$ years after injury. The fact that these patients had been seen for a period of several months overseas in the early phases of their injury gave most of the replies a personal touch which surpassed the usual casual patient-doctor relationship often encountered in the Army because of rapidity of evacuation.

TABLE 1. *Preliminary Data*

Location of Injury	Type of Injury					Associated Injury	Urinary Status								Distressing Pain	Laminectomy at 36th G.H. for Retained Bone or Metallic Fragments
	Concussion	Non-dural Penetration	Dural Penetration	?Dural Penetration	Death		Voluntary Voiding	Indwelling Catheter	Suprapubic Forward	Suprapubic at 36th G.H.	Urethrostomy Forward	Trochar	Crede ?			
Cervical—																
12 Cases	3	5	1	3	0		8	2	2	0	0	0	0	0	2 (partial) lesions	2
1–1																
2–0																
3–1																
4–4																
5–3																
6–2																
7–1																
Dorsal—																
46 Cases	3	17	19	4	3	Wounds of chest wall (30) with severe pneumothorax or hemothorax in 20 or 46.3%	6	5	15	9	4	2	0	5	3 (non-penetrating dura injuries); 1 (complete transection)	18
1–0																
2–5																
3–6																
4–4																
5–5																
6–2																
7–1																
8–4																
9–2																
10–5																
11–6																
12–6																
Lumbar—																
32 cases	3	7	18	2	2	Laceration of kidney or retroperitoneal hematoma in 8. Perforating abdominal wounds in 10 (colostomy in 7).	5	7	7	8	0	0	1	4	1 (compression of) cauda equina	14
1–8																
2–7																
3–7																
4–5																
5–5																
Sacral—																
4 cases	0	2	1	1	0		3	0	0	1	0	0	0	0		3
1–3																
2–1																
Total 94	9	31	39	10	5		22	14	24	18	4 (removed)	2	1	9		38

The questionnaire was worded simply and responses were gratifying. The descriptive replies may stem from the fact that the Army paraplegics know more about their anatomical and physiological problems than an average patient, due to long months in Army and Veteran hospitals. Follow-up on the 25 foreign cases was not possible.

Of the 55 patients who replied, 16 or 29 per cent now have no disability. It should be observed that 4 of these patients had slight to no motor disability at the time of evacuation from overseas, so that 12 patients, or 21.8 per cent, have had complete recovery from previous motor neurological disability. No improvement was shown by 20 patients who had residual neurological deficit; this is about 36.3 per cent of the entire group.

Particular reference may be made to the 19 patients who were originally completely paraplegic. Of these, 4 went on to some degree of neurological recovery. In 1, who now has a monoparesis, the dura was indented by the metallic fragment; the operator opened the dura and reported an intact but purple contused cord. In the other 3, who improved to a point of paraparesis, the conus was contused in 1; macerated cord was aspirated through the intact dura in the 2nd; and in the 3rd, the status of the cord was unknown. The remaining 15 paraplegic patients had no return of function. In 2 of these the status of the cord was not known; but in 4 others the cord was observed to be completely intact, and in a 5th, about $\frac{1}{3}$ intact. The remaining 8 patients had a complete anatomical transection of the cord.

Only 5 of the paraplegics had been made ambulatory with braces. Two other paraplegics and 2 paraparetics stated that they were unable to use braces because of their rather violent involuntary spasms. In 2 of these patients anterior rhizotomy was being anticipated for relief of spasm. A few of the patients stated that they felt the use of crutches and braces was a very definite morale booster. Another remarked that the use of crutches was excellent exercise, and he valued them from that standpoint.

One of the patients with a complete transection of the cord at the 9th dorsal vertebra had a specially equipped car for paraplegics. One afternoon he appeared for a follow-up examination, announcing that he had dressed himself, hoisted himself from his bed into a chair, rolled down a specially built incline to his garage, hauled himself up into the car dragging his col-

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Comments on Table 1:

(a) 5 patients presented spinal fluid leaks preoperatively.
(b) Mortality—American 2 deaths (2.8%).
(c) 6 patients developed severe spasms while overseas.
(d) 5 patients presented an osteomyelitis of the vertebrae—4 lumbar, 1 sacral.
(e) 17 wound infections in 94 patients (9 were in foreigners).
(f) 46 patients left 36th General Hospital with suprapubic cystotomies.
(g) Urethrostomy was found unsatisfactory.
(h) 26 patients had decubiti of varying degrees of severity.
(i) One renal calculus removed (overseas).
(j) One abscess of space of Retzius (overseas).

TABLE 2. *Follow-Up Data on 55 Patients*

Location of Injury	
Cervical	10
Dorsal	27
Lumbar	15
Sacral	3

Discharge from Service	
Within 6 mo. after injury	11 or 22%
Within 7–12 mo. after injury	18 or 36%
Within 13–18 mo. after injury	13 or 26%
Within 19–24 mo. after injury	5 or 10%
24 plus mo. after injury	3 or 6%
Still in hospital	2
Reenlisted in Army	1
No reply	2

Disability	Motor Power	Voiding Voluntarily
None	15	13
Paresis		
Monoparesis	16	13
Monoplegic		
Hemiplegia	3	3
Paraparesis	6	2
Complete paraplegia	15	—
	55	31

Bladder Status	
Voiding voluntarily	31
Indwelling catheter	3
Crede	3
Leg urinal	2
Penis clamp	3
Partial control	5
Suprapubic cystotomy	2
Questionable status	6

Pain	
None	14
Unqualified	22
Slight	13
Moderate	4
Severe	2 (3.6%)

Spasms	
None	16
Unqualified	16
Slight	7
Moderate	5
Severe	11

Comments on Table 2:

(a) 1 death had occurred, thus giving a mortality of 1.8%, $2\frac{1}{2}$–$3\frac{1}{2}$ years after injury.
(b) 6 patients had residual decubiti, 2 of which were severe.
(c) Overseas 19 of the 55 patients, or 34.5%, were voiding voluntarily. At the follow-up 31, or 56.3%, were voiding voluntarily.
(d) 26 suprapubic cystotomies were reduced to 2 (47.2% to 3.6%). 7 of this group are now voiding voluntarily.
(e) Both patients having severe pain had cordotomies (1 repeated). 1 successful.
(f) 2 patients had anterior rhizotomies and 2 were anticipating this operation. 1 had refused.
(g) 5 patients had severe spasms of abdominal muscles.

lapsed wheel-chair into the back seat, and then driven 160 miles to report on his condition.

Of the 55 patients, 24 (43.6 per cent) were actively engaged in earning all or part of their living or in furthering their education. About 30 per cent were completely supporting themselves; this includes 6 patients with monoparesis or monoplegia. Approximately 14.5 per cent were anticipating going to work; among these were 7 complete paraplegics. Two other paraplegics

TABLE 3. *Use of Mechanical Aids*

No. of Cases	Motor Status	No Motor Disability	Motor Disability without Aids	Crutches	Braces	Wheel Chair	Canes	Car
15	No disability	15						
1	" "				1 (back)			
8	Monoparesis		8					
5	"				5			
1	"				1	1		
1	"				1		1	
4	Paraparesis					4		
2	"						2	
2	Hemiplegia					2		
1	"			1		1		
8	Paraplegia					8		
3	"				3	3		
2	"			2	2	2		
1	"			1		1		
1	"					1		1
55		15	8	4	13	23	3	1

were studying a trade, and another was earning part of his living. Four patients (7.2 per cent) were taking courses of study; 2 were in college and 2 learning a trade. There were 23 men (41.8 per cent) who were contributing nothing toward their support. Only 1 patient stated, "I can't earn money." He had minor residual motor disability. Another patient with paraparesis mentioned that he was reluctant to try to earn money for fear of losing his retirement pay. This attitude may be commonly encountered. Probably more could be accomplished with these men if such fears were allayed. The occupations being performed were: Jewelry repair, photography, bookkeeping, radio repair, and one talented patient is sketching and painting pictures.

TABLE 4

No. of Cases	Degree of Disability	No Support	Anticipating Work	Studying a Trade	College	Partial Support	Complete Support
16	No disability	3	1		1	1	10
15	Monoparesis or Monoplegia	7			1	1	6
3	Hemiparesis or Hemiplegia	3					
6	Paraparesis	2	2	1			1
15	Paraplegic	8	5	1		1	
55		23	8	2	2	3	17

TABLE 5. *Progress in Motor Power*

Status at Time of Evacuation	Status at Present Time	Improved	No Improvement	Greatest Problem to Patient
4 Slight to no disability	No disability	4		21 Pain
7 Monoparesis	No disability	7		6 Loss of bladder control
5 Paraparesis	No disability	5		5 Weakness of extremities
8 Monoparesis	Monoparesis	3	5	4 Paralysis of extremities
5 Paraparesis	Monoparesis	5		3 Spasms
3 Paraparesis	Paraparesis	3		3 Stiffness of neck (cervical)
1 Paraplegia	Monoparesis	1		2 Paresthesias
3 Paraplegia	Paraparesis	3		1 Sterility
15 Paraplegia	Paraplegia		15	1 Gas on stomach
2 Triparesis	Hemiparesis	2		
1 Quadriparesis	Monoparesis	1		
1 Quadriparesis	Hemiparesis	1		
		35	20	47

CONCLUSIONS

1. A survey of 94 patients with penetrating wounds of the spine is presented. Of these, 56 patients were reviewed from the standpoint of ultimate disability and productivity.

2. The surgical management of these patients is briefly mentioned. Laminectomy was performed in wound debridement when bone or metallic fragments were present in the spinal canal. The importance of adequate roentgen studies is emphasized.

3. Of the 56 patients who could be followed, 1 had died. Sixteen (29 per cent) now have no neurologic disability. No improvement was shown in 20 patients (36.3 per cent), 15 of whom were paraplegics. Four of 19 originally complete paraplegics developed partial recovery. Five of the paraplegics have been rehabilitated to a point of ambulation with braces.

4. Forty-three per cent of the group were engaged in earning all or part of their support or furthering their education. A nearly equal percentage (41.8 per cent) contributed nothing toward their support. Of the 15 complete paraplegics, 5 were anticipating work; 1 was studying a trade; 1 contributed partial support; and 8 were completely dependent.

5. The accomplishments of rehabilitation applied to patients with spinal cord and cauda equina injuries incurred in war, deserve recognition. This philosophy of care may well be adopted in civilian injuries.

SUBDURAL EMPYEMA*

WILLIAM S. KEITH, M.B.†
Toronto, Ontario

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SUBDURAL empyema is a not infrequent intracranial complication of paranasal sinus infection. It is a much less frequent complication of otitic infection. Excellent clinical and pathological studies have been published recently by Kubik and Adams,[2] Ray and Parsons,[3] and Courville.[1] Nevertheless, reports of survivals from this disease are extraordinarily few, even since the advent of the sulphonamides and penicillin. The following 6 cases illustrate the clinical, surgical and therapeutic problems presented by this disease. Cases 1 to 4 were consecutive in the author's experience.

Case 1. W. A., male, 15 years.

On Sept. 27, 1941, 2 weeks before neurosurgical consultation, this boy sustained a crushing injury of his maxilla upward into the base of his skull. His upper teeth were resting on the top of the gate of a moving elevator. His head was caught between the car and the floor level above. He was admitted at once to hospital in Fergus, Ontario, under the care of Dr. J. H. O'Brien, where he was found to be shocked but rational.

For 2 weeks he remained rational or nearly so most of the time. He complained from the first of diplopia and some headache. Temperature varied from normal to 102, and there was occasional vomiting. He was given a lot of sulphonilamide and later sulphathiazole. He was observed to move all his limbs equally well. On Oct. 8, he seemed more drowsy and had a higher temperature. It was noted on Oct. 10 that he did not move his right foot. On Oct. 11, abnormal neurological findings were as follows. There was no voluntary power in the right lower limb except possibly slight movement at the right hip. There was sustained ankle clonus on the right and transient clonus on the left side. Plantar responses were: right—extensor, left—flexor. Tendon reflexes in all the limbs were active but were brisker on the right. The patient was right-handed but movements of the right hand were definitely slower than on the left side. Grips were equal. The left abdominal reflex was present; the right was absent. There was failure of proper convergence of the eyes. There was very slight swelling of the optic discs. There was almost no neck stiffness, but Kernig's sign was positive. There was almost no dysphasia. Sensory examination, as well as it could be carried out, was negative. C.S.F. pressure is not known. The fluid was clear. There were 4 cells and there was no growth on culture.

By the evening of Oct. 13, when he was admitted to the Toronto Western Hospital, he was very drowsy. All speech was lost except for a muttered "yes" or "no," and there was a profound weakness of the right arm.

With the worsening of his condition it was essential to establish a pathological diagnosis. Three burr-holes were made under local anaesthetic in the left frontal and temporal regions. The tension of the dura seemed normal or not much increased.

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† 208 West Block, Medical Arts Building, Toronto 5, Ontario.

There was thick, creamy, white pus over the left temporal and frontal cortex with more organization in the frontal region where the depth of pus was less. A burr-hole in the right posterior frontal region showed no sign of suppuration under the dura. Gram-positive cocci were seen and a few colonies of pneumococci were grown. He was given large amounts of sulphapyridine for several weeks. He was discharged to his home on Nov. 23, 1941, with his wounds healed and almost complete recovery of power in the right foot. His maxilla was pushed so far up in front that Dr. Fulton Risdon had to remove some posterior molars in order to get occlusion of his incisors since it was impossible to shift the impacted maxilla.

He was re-admitted to hospital on Dec. 17, 1941, with a diagnosis of left frontal lobe abscess. This abscess was drained and he was discharged on Feb. 4, 1942. He was seen in January, 1947. He has occasional Jacksonian epileptic attacks.

Case 2. G. M., male, 17 years.

On June 25, 1943, 12 days before neurological consultation, this boy complained of pain behind his left eye. The next day it was behind both eyes, worse on the left side. He reported to Dr. Scott of Hook, Hampshire, England, who found that he had some fever and a stiff neck. He was given a bottle of medicine which contained 10 gm. of sulphathiazole. He improved and wanted to return to work on July 5. Toward evening of that day, however, he had headache and vomiting and it was then found he could not stand because of weakness of the left leg.

He was sent to Park Prewett E.M.S. Hospital on July 7, under the care of Dr. Laurent. There it was found that he had fever but no neck stiffness. C.S.F. pressure was 300 mm., but fluid contained no cells; total protein was 80 mg. On July 8, he was rational, cooperated well and was well oriented. His speech was slow but clear and there was no dysphasia. He had no power in the left leg. His left arm was spastic but he could just manage with a great effort to touch his nose with his left index finger. There was a left extensor plantar response. The right abdominal reflex was normal; the left was obtained faintly. There was some lower facial weakness. There was a complete left homonymous hemianopsia and early bilateral papilloedema. The wbc. was 28,000. X-Ray plates of his skull showed the left antrum and all the ethmoid cells to be relatively opaque. The pulse had been recorded as low as 54 but it was 70 when he was examined. It was thought that he might have a septic embolus of the right anterior or middle cerebral arteries or a brain abscess in the region of the internal capsule, or a subdural empyema even though there were no cells in the C.S.F.

He was transferred to Basingstoke Neurological and Plastic Surgery Hospital. On July 8 a burr-hole was made in the right mid parietal region. There was no subdural pus. The brain bulged through the dural opening. A brain needle was passed 6 cm. toward the internal capsule without encountering any abnormality. On July 10 a brain needle was passed into the right temporal lobe. No abscess was found. The needle entered the temporal horn of the ventricle and oxygen was put in. There was no significant displacement of, or changes in the ventricular system by this rather inadequate ventriculogram.

His condition deteriorated day by day with increasing drowsiness, paresis, papilloedema and bilateral extensor plantar responses. At this stage, Dr. E. H. Botterell suggested that one more attempt be made to find pus. The only remaining spot to attack was the right occipital region. On July 21 a brain needle was passed through the right occipital lobe. When it neared the falx it entered a large abscess cavity

About 30 cc. of pus were removed and 10 cc. of thorotrast were placed in the cavity. X-Ray (Fig. 1) showed a subdural abscess around the right occipital pole and along the falx for $\frac{2}{3}$ of its length. This abscess was drained on the following day. There was no growth by aerobic or anaerobic cultures and no organisms were seen in direct smear. The patient made a gradual but steady recovery. By the end of August, over 1 month after the abscess was drained, he was talking normally. He walked with care and not quite normal speed. He had a partial left lower homonymous visual field defect. His limbs were still slightly spastic with some clonus at the knees and ankles and increased reflexes. The left leg was weaker than the right. The small occipital skull defect was soft, pulsating, and there was no bulging. Sulphadiazine was

FIG. 1. Case 2. Thorotrast demonstrates abscess around right occipital pole and along falx 4 weeks after onset of illness.

administered in the usual therapeutic doses through the course of his illness and until shortly before his discharge from hospital. He was discharged to Park Prewett Hospital, Basingstoke, Hampshire, and shortly thereafter returned to his home and regular employment.

Case 3. G. B., male, 38 years.

It was noted on this man's entry into the R.C.A.F. that he was subject to chronic bronchitis. On Sept. 25, 1943, abdominal pain developed and a diagnosis of subacute appendicitis was made. The appendix was removed on Sept. 30, 1943. Following operation there was a swinging temperature up to 101. For the following month he had intermittent fever, cough, purulent sputum, and pain in the right side of the chest. Administration of sulphathiazole diminished the chest symptoms but they recurred as soon as the drug was stopped. It was thought that he had bronchitis followed by infarction of the lung as the sputum became blood-tinged. He had a right-sided pleural effusion which on aspiration showed lymphocytes but few polymorphs. A gram-negative bacillus and some anthracoid bacilli were grown. On Nov. 10, 1943, he was admitted to No. 7 Canadian General Hospital, Taplow, England.

A diagnosis of lung abscess was made. He was certified unfit for further service and remained ill with productive cough and probably some bronchiectasis until about Feb. 20, 1944, when red tender swellings developed in the scalp of the left frontal and right occipital regions. He was given sulphadiazine. The swelling subsided but a slight fever persisted. Sulphadiazine was discontinued on Feb. 29, 1944.

About this time the nurse's notes indicated gradually increasing headache, irritability and some drowsiness. On Mar. 4, 1944, he had severe left frontal headache and vomiting. This was followed later in the day by severe dysphasia. He was then transferred to Basingstoke Neurological and Plastic Surgery Hospital.

On examination he was very dysphasic but occasionally replied to questions with a single word which was not always intelligible or relevant. Repeated movements of the hands were somewhat slower on the right than on the left. There was no neck stiffness. C.S.F. pressure was 250 mm. The fluid contained 110 polymorphs and 30 lymphocytes per c.mm. Wbc. was 14,000. There was some lower right facial weakness. Diagnosis was left frontal abscess with possible involvement of temporal and parietal lobes. The possibility of subdural empyema was considered. The lesion was considered to be secondary to or associated with an unusual and rather mild osteomyelitis of the skull.

On Mar. 5, 1944, a burr-hole was made under local anaesthetic in the left frontal region. The surface of the frontal bone was rough and granular. Pus was found under the arachnoid. This pus was followed deep into the left frontal lobe. There seemed to be very little inflammatory reaction in the brain tissue itself. A small tube was left in the depth of the purulent focus. The direct smear showed large numbers of streptococci. On culture, the organism proved to be a non-haemolytic micro-aerophilic streptococcus which was penicillin sensitive. The patient had several right-sided seizures in the first 2 days after operation. These seizures were followed by a mild right hemiparesis which gradually cleared up in about 10 days. On Mar. 7 the frontal lobe was again explored. More pus was found in front of the tube deep in the brain. Subdural pus was found over the lateral surface of the left frontal and temporal lobes. There was no subdural pus in the left parietal region or in the right frontal region. The patient became very drowsy and had persistent vomiting. Further exploration of the frontal and temporal lobes failed to locate any more pus. He was given penicillin intramuscularly, intrathecally and also directly into the wound by means of a fine catheter. The intrathecal injections were stopped after the 3rd day.

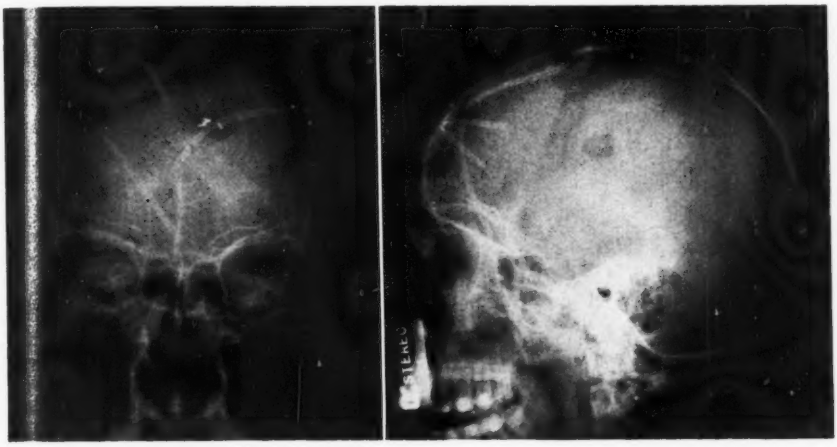
He slowly recovered from the signs of local damage to his brain and from the signs of swelling of the brain. But his wounds repeatedly broke down. Further X-Rays of his skull then showed a fairly extensive left frontal osteomyelitis. On May 4, 1944, a large block of left frontal bone was removed. Shortly afterward his wounds healed and he was returned to Canada.

In Christie Street Hospital, Toronto, he was under the care of Dr. K. G. McKenzie and Dr. C. P. McCormick for recurring osteomyelitis of the skull. He had a further resection of frontal and temporal bone and a great deal more penicillin. In September, 1944, he was discharged to his home with his wounds healed and his brain functioning normally.

Case 4. C. A., male, 24 years.

Early in March, 1944, this patient had a head cold. On Mar. 14 he had pain above the right eye. This was later accompanied by swelling and tenderness around the right eye and across the forehead. On Mar. 18, 1944, he was admitted to No. 9

Canadian General Hospital, Horsham, Sussex. On Mar. 20 a diagnosis of acute right frontal sinusitis with cellulitis of the forehead was made and on Mar. 21 Major G. A. Henry, R.C.A.M.C. made an opening in the right eyebrow. This led to a hole in the frontal bone through which a large amount of pus escaped from the right frontal sinus. There was no exudate in the nose. He was treated with sulphathiazole. On Mar. 25 he had a convulsion involving the left arm and he was immediately transferred to Basingstoke Neurological and Plastic Surgery Hospital. A second seizure was observed on that day. It began in the left hand and spread to the trunk, leg, face and then to a limited extent to the right face and hand. Wbc. was 23,000. C.S.F.



FIGS. 2 and 3. Case 4. Catheters in subdural space near falx for local penicillin therapy.

pressure was 250. There were 17 polymorphonuclears and 6 lymphocytes in the fluid. The diagnosis was subdural empyema, right side.

Operation was carried out on Mar. 25 under local anaesthetic. A burr-hole was made in the left frontal region close to the midline. Tension of the dura seemed normal. On opening the dura, a bead of pus formed. This was traced to the space between the frontal lobe and the falx. A fine catheter was placed in this space against the falx. Two burr-holes were then made to the right of the midline, in posterior frontal and mid-parietal regions. Subdural pus was encountered in each of these, coming more from midline and forward rather than lateral and behind. The brain was not greatly swollen. Catheters were passed down to the falx through each burr-hole. An abscess involving the right frontal region and right orbit was then drained. The patient was treated by intramuscular, intrathecal and subdural routes with penicillin (Figs. 2 and 3).

A streptococcus was seen in direct smear. It proved to be non-hemolytic, microaerophilic and penicillin-sensitive. He made an apparently complete recovery in less than 3 weeks. Air encephalography was done on April 25, 1944. The ventricles and subarachnoid air markings were normal.

On Dec. 1, 1944, he reported to Christie Street Hospital, Toronto, because of frequent headaches over the right eye. His right antrum was found to contain foul

pus. It was washed out two or three times and finally he had intranasal drainage by Dr. Howard McCart on Jan. 3, 1945.

Case 5. A. F., male, 20 years.

This young man had tonsillectomy performed on Nov. 12, 1945. Recovery was uneventful. On Nov. 22 there was pain and swelling over the right eye. On Nov. 26 he became severely ill, drowsy and mentally confused. Lumbar puncture was done because of neck rigidity. The spinal fluid was clear and the pressure was said to be normal. The diagnosis at that time was acute frontal sinusitis with beginning abscess formation involving the bone and meninges. He was given sulphadiazine and penicillin and his condition improved slightly for 2 days. On Nov. 29 the patient became more drowsy and on that and the following day he had 15 or 20 seizures involving the whole right side of the body. On Nov. 30 an abscess in the frontal region between the skin and the bone was drained.

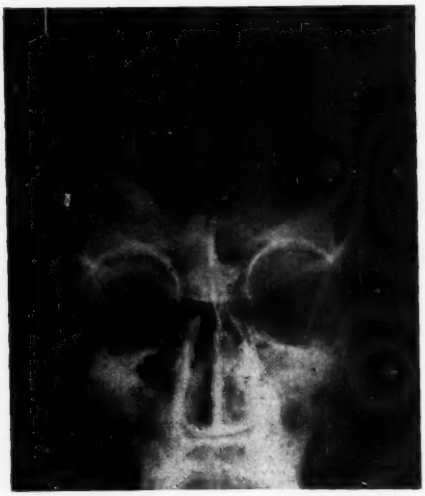


FIG. 4. Case 5. Shows extent of removal of bone. Operation was done by Dr. K. G. McKenzie.

On Dec. 1, 1945, he was admitted to the Toronto General Hospital for neurosurgical treatment. He was drowsy and looked toxic. There was moderate neck rigidity. He responded fairly well to questions and stimuli. There was marked choking of the left optic disc; the right was not seen. There was weakness of the whole right lower limb with paralysis of the muscles of the calf and the dorsi-flexors of the ankle. The tendon reflexes of the right leg were very active; there was clonus at the ankle, and the plantar response was extensor. There had also been some twitching of the left side of the body. The diagnoses were thrombosis of the superior longitudinal sinus or subdural empyema.

Operation was done under local anesthesia on Dec. 2, 1945. Subdural pus was found through a burr-hole in the upper left rolandic region. Left mid temporal and occipital burr-holes showed normal-looking arachnoid and brain. It was then decided that drainage would be unsatisfactory through burr-holes so a block of bone, $2\frac{1}{2}$ inches wide, centering on the longitudinal sinus was removed from the frontal sinuses to the rolandic region (Fig. 4). Both frontal sinuses were widely opened. Anterior and posterior walls were removed. Both sinuses contained pus with extradural pus behind the sinuses. Subdural pus was found on both sides of the superior longitudinal sinus and falx back to the rolandic area. Soft rubber drains were placed between the brain and the falx on either side and under the tip of each frontal lobe. The patient was treated with sulphadiazine by mouth and the subdural space was irrigated with penicillin every 4 hours. He was also given 15,000 units of penicillin every 3 hours by continuous intramuscular drip. On 3 occasions of 2 or 3 day intervals, he was given 20,000 units of penicillin intrathecally by os-

ternal puncture. The paresis of the right leg and foot cleared up completely within 2 weeks. Examination of the pus from the subdural empyema showed a few gram-positive cocci in pairs and on culture, haemolytic staphylococcus aureus was grown.

On Dec. 9, 1946, he looked very well. He had gained more than 20 pounds in weight since before his illness. There was a large soft defect in the mid-frontal region with an island of new bone floating in the middle of it. He had no complaints.

Case 6. J. R., male, 10 years.

On Dec. 11, 1945, this boy complained of headache above his left eye but had no fever. On Dec. 13 his temperature was 103 and he had severe pains in the right frontal region. On Dec. 17, temperature was 104 and the pain had spread into the right temple. He was given regular doses of sulphathiazole followed by a drop in temperature. When he began to vomit, the sulpha drug was discontinued. On Dec. 19 he became stuporous. He was admitted to the Hamilton General Hospital where he received regular doses of penicillin intramuscularly and sulphathiazole by mouth.

The patient remained semi-stuporous, and probably had his first seizure on Dec. 24. On Dec. 25 he was transferred to the Toronto General Hospital and while being examined he had a seizure involving first the left leg and then the left arm and face. It did not become generalized. On or about Dec. 23 there were 80 to 100 cells in the spinal fluid and he was given penicillin intrathecally. On the same day his wbc. was 12,000, on Dec. 25, 40,000, and on Dec. 26, 28,000.

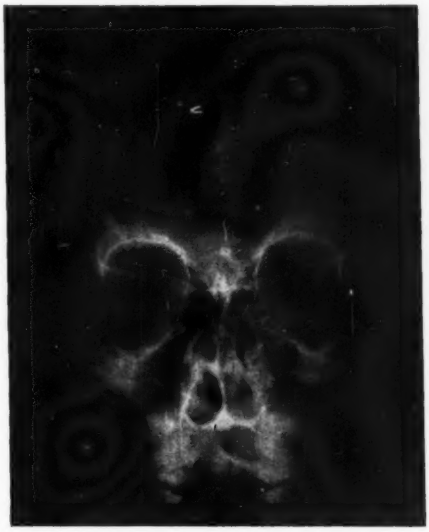


FIG. 5. Case 6. X-ray showing defect in frontal region 9 months after operation. Dr. E. H. Botterell's patient.

On Dec. 26 X-Rays of the skull were negative except for cloudiness of the accessory air sinuses. He was restless and responded to painful stimuli. There was no papilloedema. The left side of the face moved normally but the left leg showed a considerable degree of spastic paralysis. The left arm was not examined because of extensive thrombophlebitis. The plantar response on the left side was extensor and on the right side it was equivocal. The clinical diagnosis was subdural empyema right and probably left, secondary to acute frontal sinusitis.

Operation was done on Dec. 26 under local anaesthetic with the patient in a semi-sitting position. Through a midline incision in the scalp, the right frontal sinus was removed and an extradural abscess behind it was evacuated. Subdural pus was found to a depth of $\frac{1}{2}$ inch over the right frontal pole and extending to the medial side of the hemisphere. A channel was then cut in the skull, back to the pre-rolandic area (Fig. 5). The dura was opened throughout its length and purulent exudate was found between the brain and the falx. Drains were placed on the medial and lateral surfaces of the hemisphere and also on its orbital surface. On the left side, the left

TABLE 1

Case	Sex and Age	Days of Illness Before Consultation	Symptoms of Illness	Early Therapy	Neurological Symptoms	Pathological Findings	Treatment	Organism
1	W.A. Male 15	14	Fracture through maxilla	S'nylamide and S'thiazole	Increasing drowsiness. Weakness right leg.	Left fronto-temp. Subdural empyema. Later L. frontal abscess.	3 burr-holes, 1 negative burr-hole right side. S'pyradine.	A few colonies of pneumococcus
2	G.M. Male 17	12	Pain behind eyes. Stiff neck.	S'thiazole	Weakness left leg. Later hemiplegia, hemianopsia.	Empyema medial side of right hemisphere and around right occipital lobe.	Negative burr-holes and brain punctures. S'diazine. Then drained through occiput.	No growth aerobic or anaerobic.
3	G.B. Male 38	13 (4 mos. illness. Lung abscess?)	Swelling of scalp. Headache. Vomiting.	S'diazine	Dysphasia & postop. Jacksonian seizures.	Abscess left frontal lobe. Left subdural empyema. Osteomyelitis skull.	Repeated burr-holes. Penicillin. Later removal of portions of skull.	Micro aerophilic. Non-haemolytic. Streptococcus. Penicillin sensitive.
4	C.A. Male 22	10	Pain and swelling over and around right eye.	S'diazine	Jacksonian seizures. Delirium.	Subdural empyema. Ant. part falx and rt. frontal.	Burr-holes and penicillin.	
5	A.F. Male 20	9	Pain and swelling over rt. eye. Drowsy, confused, stiff neck.	S'diazine and penicillin.	Jacksonian seizures. Weakness rt. leg.	Subdural empyema both sides. Back to rolandic region. Frontal sinusitis. Extradural pus.	Removal of block of bone mid frontal. Penicillin, sulphadiazine.	Haemolytic. Staphylococcus aureus.
6	J.R. Male 10	14	Pain frontal. High fever. Stupor.	S'thiazole. Later penicillin.	Jacksonian seizures, starting in left leg. Spastic paresis of left leg.	Frontal sinusitis. Extradural abscess. Subdural empyema. Right frontal and parasagittal.	Channel cut in skull from frontal sinus to rolandic region. Penicillin.	No organisms seen, and no growth on culture.

frontal sinus was removed, extradural pus was found behind it; subdural pus was not found but infra-orbital and parasagittal drains were placed for the instillation of penicillin. Right temporal and occipital burr-holes showed no evidence of subdural pus.

Repeated examinations of pus from extradural and subdural spaces did not reveal any bacteria in the direct smear and there was no growth on culture.

The patient was treated with penicillin intramuscularly, intrathecally and by local irrigation in the tubes. He was fed by duodenal tube and he was given phenobarbital. By Dec. 29 he could drink normally; he was no longer incontinent and phenobarbital was discontinued. By Jan. 7 he was talking well, temperature was normal, and penicillin was discontinued. He had a total of 3,000,000 units of penicillin. He was discharged to his home in good condition on Jan. 20, 1946.

On June 24, 1946, he looked to be in excellent health. There was a bone defect in the mid-frontal region which pulsated. There had been no fits or spells of any sort. Neurological examination was negative except for some increase in tendon reflexes in the left leg.

DISCUSSION

Case 3 is rather different from the others, both clinically and pathologically. There was a subdural empyema over the lateral surfaces of the frontal and temporal lobes but there was also pus within the frontal lobe and there was fairly extensive osteomyelitis of the skull.

In the other 5 cases the infection appears to have entered the subdural space by way of the paranasal sinuses. In Case 1, trauma provided the means of entry. In Case 2 it was not established that there was any sinusitis other than by the appearance of the X-ray plates. In Cases 4, 5 and 6, the infection spread from the frontal sinuses. It is worth the attention of anyone interested in neurological diagnosis or in the surgical treatment of this disease that in 4 of these 5 cases there is good evidence that the leg area was involved in the suppurative process before the arm area. This is probably due to the spread of pus up one or both sides of the falx. If the author had been aware of this in Case 1, more adequate drainage at the first operation might have led to a more prompt recovery, avoidance of frontal lobe abscess and his present infrequent epileptiform attacks. It was because of this observation that the parasagittal region was so carefully explored, beginning on the less suspected side in Case 4. This knowledge was of service also to Dr. McKenzie and Dr. Botterell in their operations in Cases 5 and 6.

In 42 autopsies showing subdural empyema, Courville[1] found pus on the medial side of the cerebral hemisphere on 4 occasions. On the other hand, Ray and Parsons[3] in 2 out of 3 autopsies found that pus had extended under the falx from the major lesion on one side to involve the subdural space on the other side. Kubik and Adams[2] state: "The subdural exudate generally covered a large part of one cerebral hemisphere. The pus was most abundant in the lower lateral frontal region, anterior to the motor cortex and as a rule extended to the medial and orbital surfaces, to the temporal lobe and as far back as the parietal or posterior parietal region. In some cases it extended beneath the falx to the opposite hemisphere. . . . Beneath the subdural

exudate the lateral surface of the frontal lobe was always depressed, as in subdural haematoma."

In the first 2 cases sulphonamides only were used. Penicillin was not then available. In each case the history makes it fairly certain that the sulphonamide therapy modified the course of the sepsis to such an extent that the patient survived until a local lesion formed, was diagnosed and could then be treated surgically. It is likely that in each of the 4 latter cases the sulphonamides had at least a favourable influence on the progress of the infection. Early treatment of subdural empyema and its precursors with sulphonamides is likely to occur almost always nowadays. Sulphonamide therapy may prevent the development of subdural empyema in some cases and may modify the more severe cases so that they can be treated by operation and penicillin as well.

Acute suppurating lesions about the head and particularly those of the paranasal sinuses should be treated vigorously with sulphadiazine and penicillin to prevent intracranial extension. When the diagnosis of subdural empyema is suspected, the matter should be settled at once by making a burr-hole in the suspected region, usually posterior frontal, 2 or $2\frac{1}{2}$ inches from the midline and a second burr-hole close to the falx in the frontal region. Both sides should be explored if necessary. If subdural empyema is found, radical surgical drainage by removal of bone and opening of the dura is indicated. Pus should be looked for on all surfaces of the frontal lobes. Both penicillin and sulphonamides should be used to supplement the surgical treatment.

SUMMARY

Six cases of subdural empyema are reported.* All the patients survived the disease. Sulphonamides were used in all cases. Operation for diagnosis and drainage was done in each case. Penicillin was used in all but the first 2 cases. The relationship between weakness of one leg and empyema on the medial side of the other cerebral hemisphere is noted and discussed.

NOTE BY K. G. McKENZIE

Up until a few years ago most neurosurgeons felt that subdural empyema was a hopeless disease. Some years ago a patient of Doctor Kahn was cured by drainage through burr-holes associated with frequent and massive lumbar puncture to keep the brain away from the dura and so favour adequate drainage. Subsequent to seeing this case I treated several patients in a similar manner but was unsuccessful. Always the brain swelled and prevented adequate drainage through burr-holes. My first success was in Case 5 (reported by Dr. Keith). Since that case there have been 3 more recoveries in this centre. All of these patients were dreadfully ill. Undoubtedly their recovery is in the largest measure due to penicillin but in part we think due

* The author is deeply indebted to Dr. K. G. McKenzie and Dr. E. H. Botterell of the Neurosurgical Service, Toronto General Hospital, for their courtesy in allowing publication of their cases, 5 and 6 respectively.

to one or two technical points which may be of interest to other neurosurgeons.

Placement of soft Penrose drainage tubes in all areas where there was subdural pus was best accomplished by taking out a block of bone along the midline from the frontal sinuses to the pre-Rolandic area; at the same time the anterior and posterior walls of the infected frontal sinuses were removed. This permitted easy exploration of both sides and adequate flushing out of the whole infected field. Drainage through the flattened-out soft tubes was nil and not a factor in the recovery. The essential principle was an arrangement whereby penicillin could be injected into a space which normally is not flooded by systemic or intrathecal penicillin. (This is assumed to be so but is not definitely proved.) Incisions were closed around the soft drainage tubes, a dressing was applied with the drainage tubes projecting through the dressing, and a short length of firm rubber tubing which fitted a syringe was then tied in to each of the projecting Penrose drains. Each tube was then plugged with a small wooden peg (golf tee), a second dressing was then applied covering in all the tubes. Subsequently over a period of 3 or 4 days penicillin was injected through each tube after removal only of the outer dressing; the wooden pegs prevented the escape of the penicillin; the soft drainage tubes do not damage the brain. The inner dressing was not removed until stitches and the tubes were removed, usually on the 4th or 5th day. In no case did any secondary infection develop in the incisions.

The 6 cases that Doctor Keith reported, all patients having recovered, mark a new era in surgical treatment of intracranial infection. In addition 3 others could be added from this centre to bring the total up to 9.

ADDENDUM

It was felt worthwhile to add the following case report. The patient's illness occurred after this paper was prepared for publication.

Case 7. M. M., male, 17 yrs.

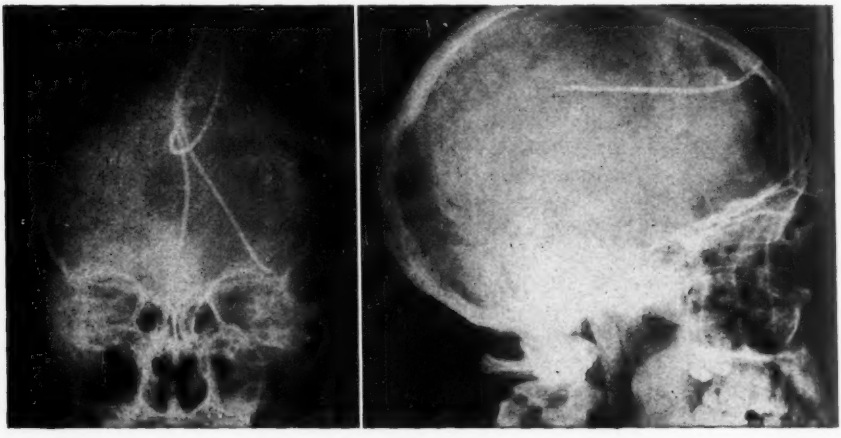
About Jan. 18, 1948, this young man began to have pain in the right side of face and forehead. He remained in bed at home while the pain increased. His temperature was as high as 102. He was given sulphadiazine by mouth, but because of vomiting this was changed to large doses of penicillin by mouth. The vomiting continued. There was no swelling of the face and forehead. On the afternoon of Jan. 23, he complained of his left foot being weak and dragging a little. On Jan. 24, he was unable to move the foot upward and quite unable to move the toes.

On Jan. 24, his temperature was 100.5 and pulse was 80. Apart from slight acne there was no appearance of inflammation or swelling of the face or forehead. There was slight tenderness over the right antrum and over the right frontal sinus and most of the right forehead. The tenderness extended on the right side up to about the coronal suture, and was much more marked near the midline than toward the temporal region. There was very slight neck stiffness. With great effort and after some delay the patient could plantar-flex the left foot. He moved the left hip and knee well. The plantar response was very slight and mostly flexion. There was transient clonus at the left ankle. Sensory examination of both lower limbs was

normal. The rest of the neurological examination was negative except for some very slight weakness of the left arm as compared to the right.

The clinical diagnosis was subdural empyema from the right frontal sinus or the ethmoid cells. Wbc. was 17,000. C.S.F. pressure was 140 mm. The fluid was clear. There were 31 cells. X-Rays of the skull showed no evidence of osteomyelitis. The right antrum and frontal sinus were cloudy, while those on the left side appeared normal.

On the evening of Jan. 24, under general anaesthetic, a burr-hole was made at the coronal suture to the right of the midline. The diploe appeared to exude pus. A nick was made in the dura and pus flowed out freely. The forehead was then scalped and bone was removed from this point forward to the right frontal sinus. Most of the right frontal sinus was also removed. The dura was then opened well forward to the



FIGS. 6 and 7. Case 7. Shows urethral catheters in place for local penicillin therapy.

right of the midline. Pus was found on the medial surface of the right hemisphere, and over the outer surface of the frontal lobe. There was less pus on the orbital plate. Exploration backwards showed that there was no pus on the lateral surface of the brain in the rolandic region, but there was a great deal of pus between the brain and the falx backwards to perhaps a little more than half way from the frontal to the occipital region. The dura over the left frontal pole was then exposed near the midline. When it was opened no pus was found over the surface of the left frontal lobe, or between the brain and the falx. Three pieces of urethral catheter were then placed in the subdural space against the falx on the orbital plate and behind the right frontal sinus (Figs. 6 and 7).

Four cultures were taken from different places. Aerobic and anaerobic cultures were made. There was scanty growth of staphylococcus aureus from one subdural specimen only. The patient was given penicillin systemically and through the urethral catheters. He was also given sulphadiazine. About 40 hours after operation he had a generalized convulsive seizure, in spite of the fact that he had been having phenobarbital gr. 1 every 4 hours. This was the only convulsion in the course of his illness.

From this point his convalescence was uneventful except for moderate drowsiness, intermittent headache, vomiting on one or two occasions, and considerable bulging through the bone defect at times.

He was able to dorsiflex his left foot 48 hours after operation, and on the following day he could move his toes, including independent movement of the great toe. During convalescence he was given a course of staphylococcus toxoid. Six weeks after the onset of his illness there was no trace of weakness in his left foot, and he appeared to have made a complete recovery.

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PHYSIOLOGICAL SALT SOLUTIONS FOR BRAIN SURGERY

STUDIES OF LOCAL pH AND PIAL VESSEL REACTIONS TO BUFFERED AND UNBUFFERED ISOTONIC SOLUTIONS

K. A. C. ELLIOTT, PH.D., AND H. H. JASPER, M.D.*

Department of Neurology and Neurosurgery, McGill University, and the Montreal Neurological Institute, Montreal, Canada

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IT IS common experience that simple exposure of the pia-arachnoid surface of the cerebral cortex during prolonged neurosurgical procedures, such as for an exploratory craniotomy, results in the formation of adhesions between the dura and arachnoid. In certain cases there may be transitory aphasia or paresis during the first week following a simple exploratory procedure. These transitory postoperative complications are usually attributed to "edema." This explanation has received support in the experiments of Prados, Strowger, and Feindel,[15,16] who demonstrated that marked edema does develop following simple exposure of the cortex in cats and that it is associated with vasodilatation and an increase in permeability of the blood-brain barrier. They found that this reaction of the brain to exposure could be prevented, at least in part, by hormones (desoxycorticosterone, adrenal cortical extract) which tend to counteract the increase in vascular permeability.

The physical factors (temperature, mild trauma, air, light, drying, etc.) acting upon the exposed brain to produce these undesirable reactions have not received systematic study. One of these factors may be the composition of the irrigation fluid. The study of irrigation fluids for use on the exposed brain, which forms the subject of the present report, is a part of a series of investigations on the general problem of the reaction of the brain to exposure which is being carried out under the direction of Professor Wilder Penfield.

The importance of the composition of salt solutions in experimental studies of the nervous system has long been recognized.[2,9,12] Studies of the *in vitro* metabolism of brain tissue[4,7] have shown that it is sensitive to the composition of the medium in which the tissue is bathed. Much recent work has shown the dependence of the physical condition of biological materials, and the activity of many enzymes, upon the presence of various ions, often in low concentration. Little consideration has been given to the possible significance of these findings in the selection of solutions most suitable for use as irrigation fluids during operations on the brain or elsewhere or for replacing spinal fluid or filling cavities. There is little uniformity of practice among neurosurgeons with regard to the type of solution employed except that it is usually made isotonic with serum. Some use normal saline (0.85 or

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0.9 per cent NaCl) while others use other "Ringer" solutions of varying composition.

Hartmann[10] (see also Sachs[18]) has described a solution resembling spinal fluid particularly in its content of bicarbonate, but this was apparently used only for filling the ventricles of hydrocephalic infants after drainage for electrocoagulation of the choroid lexus. In spinal fluid, and in all other body fluids, the concentration of the bicarbonate ion is exceeded only by those of sodium and chloride; it constitutes about 15 per cent of the total anionic concentration. Besides serving as the main physiological buffer it is concerned in other biochemical mechanisms. Any solution that lacks this ion is therefore highly unphysiological.

In the present communication the preparation and dispensation of sterile fluids closely resembling cerebrospinal fluid are described, general methods for the study of the effects of various fluids are outlined, and studies of the reaction of the cortex to the pH of irrigating solutions are reported. Studies on the effects of different irrigation fluids on the electrical activity of the cortex, the response to cortical stimulation, the development of edema, and the formation of adhesions and other aspects of the problem are in progress.

PREPARATION OF STERILE IRRIGATION FLUIDS

The preparation of two solutions resembling spinal fluid has been worked out so that technicians would not be called upon to weigh fractions of grams, and manipulations are reduced to a minimum. The first of these, Solution A, is very similar to Locke's or Tyrode's solution. It lacks bicarbonate but is readily prepared. The second, Solution B, contains bicarbonate and is somewhat more troublesome to prepare in sterile condition. It resembles the solution designed by Hartmann[10,18] but is more easily prepared in bulk and approaches the composition of spinal fluid a little more closely. In Table 1 the composition of these solutions is compared with that of cerebrospinal fluid and blood plasma.

Though the above solutions were designed with operations on the brain in mind, they should theoretically be suitable for irrigation of other organs and tissues and as perfusion fluids in experimental physiology. They differ from commonly used Ringer solutions in having a slightly lower content of calcium but the amount present is approximately equal to that in spinal fluid and to the free, ionized, calcium of serum.

Solution A. This solution is prepared according to the following simple formula and may be autoclaved. It should be autoclaved immediately after preparation.

NaCl	86	g.
KCl	3	g.
$CaCl_2 \cdot 2H_2O$	2	g.
$MgSO_4 \cdot 7H_2O$	3	g.
Dextrose	10	g.
Distilled water	10.5	liters*

This solution contains no phosphate since this ion causes precipitation of calcium

* Theoretically the volume should be 10 liters but approximate allowance is made for the amount commonly lost by evaporation during the autoclaving of 3 liter quantities.

and magnesium during autoclaving. Like distilled water or any unbuffered solution of neutral salts, Solution A after standing usually has a slightly acid reaction, about pH 6, due to dissolved carbon dioxide.

This solution has been adopted for all operative work in the physiological laboratories of the Montreal Neurological Institute. The same solution without glucose is used routinely in the operating rooms of this Institute. For the present, glucose is omitted since immediate autoclaving of large quantities cannot always be insured and the value of the glucose has not yet been proven.

*Solution B.** Sterile preparation of this fluid requires the preparation of three separate solutions.

Alkaline Salt Solution
Stock Solution

NaCl	236 g.
Na_2CO_3	92 g.
KCl	12 g.
$Na_2HPO_4 \cdot 12H_2O$	8 g.
Phenol red 1 mg/ml	4 ml.

Dissolved, filtered through sintered glass, and made to 1 liter with distilled water.

Use Solution

25 ml. of the stock solution are diluted with distilled water to 1 liter.

Acid Salt Solution

$CaCl_2$	24 g.
$MgCl_2 \cdot 6H_2O$	12 g.
Concentrated HCl	300 ml.

Glucose Solution

Dextrose (anhydrous)	8 g.
Water	500 ml.

Pyrogen-free water should, of course, be used throughout. The high salt concentration and strong alkalinity of the "stock alkaline salt solution" prevents the growth of organisms. This solution may therefore be kept for a long time provided that it is well stoppered. It should be kept in a pyrex bottle. The phenol red solution is prepared by dissolving 100 mg. phenol red (phenol sulphonephthalein) in 5 ml. 0.1 N NaOH and diluting to 100 ml. with strong sodium chloride solution, 20 g. per 100 ml.

The acid-salt solution, after gentle warming to dissolve the magnesium salt, is filtered through a sintered glass filter with gentle suction and stored in a bottle with a helmet screw cap. It cannot be autoclaved but is self-sterile. It needs to be prepared only occasionally since it is stable (if well capped) and one 300 ml. preparation is sufficient for nearly 150 liters of mixture. This solution should, at least initially, be prepared by a qualified chemist. The bottle should be stored away from surgical instruments or kept in a covered specimen jar.

The alkaline salt and glucose solutions are autoclaved in convenient quantities, usually 3 liter and 150 ml. lots respectively. Acid-salt solution is added** to the alkaline salt solution—a little more than 2 ml. of the former per liter of the latter—with thorough mixing, until the pink color† is discharged. The sterile glucose solution, 50 ml. per liter, is added and the whole well agitated. The mixture should be

* Some of our neurosurgical colleagues call this "artificial spinal fluid."

** This addition is best made by means of a sterile syringe with a thin glass tube, instead of a needle, attached by rubber tubing. In view of the strong buffering capacity of the bicarbonate, exact measurement of the acid solution is not critical.

† The strong pink color of the alkaline salt solution makes it unlikely that un-neutralized fluid would ever be used by mistake.

prepared shortly before use; it should not be warmed above about 55°C. and should not be exposed in an open container for long periods.

The following facts necessitate the above method for preparation of Solution B. A bicarbonate solution cannot be sterilized by heat since the solution would become strongly alkaline as carbon dioxide is lost and the bicarbonate is converted to carbonate. Seitz filtration would not be practically feasible for routine preparation of large volumes of solution. The amount of concentrated acid added is just sufficient to convert the carbonate to bicarbonate and to produce sufficient carbonic acid, dissolved CO_2, in the solution to give a nearly physiological pH. Calcium and magnesium salts would be precipitated by the carbonate solution and are therefore dissolved in the acid. The calcium chloride is readily soluble in the acid. But magnesium chloride is not very soluble and the amount of magnesium introduced in this way is not quite as high as the average amount in spinal fluid. Magnesium sulfate cannot be used since its addition to the acid solution causes precipitation of calcium sulfate. A trace of sodium sulfate could be added to the alkaline salt solution but there is no reason to believe that sulfate ion is essential. Sterile glucose solution has to be prepared separately since it becomes caramelized on autoclaving in the alkaline carbonate solution. The volume of the glucose solution approximately compensates for the loss of volume of the main solution during autoclaving.

TABLE 1

Composition of fluids mg. per 100 ml.

	Blood† Plasma	Spinal Fluid*†		Solution B	Hartmann's Solution	Solution A	Ringer-Locke	Ringer Tyrode
		Average	Range					
Chloride (as NaCl)	594	726	700–750	762	806	900	958	849
Bicarbonate (as $NaHCO_3$)	193	177		182	198	—	10–30	10
Phosphate (as P)	4	1.5	1.2–2.1	1.7	—	—	—	1.3
Sulfate (as S)	3.4	?	?	—	—	3.9	—	—
Sodium	316	324	301–343	335	345	338	360	318
Potassium	19	13	11–15	15.8	20	15.8	22	10.5
Calcium	10	5	4.5–5.5	5.5	5.5	5.5	8.7	7.2
Magnesium	2	3	1.0–3.5	0.9	4.8	2.9	—	2.6
Glucose	100	61	50–80	80	—	80	(100)	100
Freezing point	−0.57	−0.57	−0.53 to −0.6	−0.53‡	−0.55‡	−0.54‡	−0.58‡	−0.51‡
pH	7.4	—	7.35–7.40	7.3±	7.3±	6.0±	7.9–8.3§	7.4§

* Figures given by Merritt and Fremont-Smith.[12]

† Blood plasma contains, of course, some 7 per cent of proteins and normal spinal fluid contains a trace of protein. Both contain small amounts of urea, uric acid, amino acids, creatinine and other materials.

‡ Calculated values.

§ The amount of bicarbonate present in these solutions is too low to provide appreciable buffering capacity and the pH would fall considerably in the presence of CO_2.

Solution B has an initial pH of about 7.3. On standing exposed, or if heated, the solution loses carbon dioxide and becomes alkaline and calcium and magnesium carbonates precipitate. In an open dish at about 37°C., the pH rises gradually to about 7.8, the solution becomes noticeably pink, and a faint scum of precipitate forms, usually within about 2 hours. (The pink color is apparent before excessive alkalinity has developed.)

Solution B has been subjected to tests on animals and is now being used in this Institute on a trial basis as irrigation fluid in operations involving craniotomy and as replacement fluid after ventriculograms and after removal of neoplasms. Results of these trials will be reported later.

Methods for dispensing irrigation fluid for operations have been designed to avoid prolonged exposure and to maintain the fluid at a reasonably constant temperature without the danger of inadvertent overheating. In the simplest method the irrigation solution is kept in a welded stainless steel tank which stands in a serological constant temperature water bath. A 150°C. thermometer, fixed by a rubber stopper through a hole in the top of the tank, gives the temperature of the bath fluid. The tank is covered by a stainless steel lid which is left in place when the fluid is not in immediate use. (Holes may be cut in the lid large enough to hold inverted rubber bulb irrigation syringes so that the spouts dip deeply into the solution. These holes are covered by hinged lids when no bulb is in place.) The metal of the tank is extended as an "apron" round the bath in order to prevent contamination of sterile clothing by chance contact with the bath. The tank and thermometer with rubber stopper, are sterilized by autoclaving. The bath is filled to a level which will not allow overflow when the tank is in position. The water in the bath contains 2 per cent "Lyso-septol" as a safety factor and a strong dye so that if a leak should ever develop in the tank, the entry of bath fluid would be immediately detected. The apparatus now in use (Fig. 1) consists of twin tanks, made in one piece but with 2 inches space between them. This allows fresh solution to warm up on one side while the solution on the other side is being used.

Another apparatus,* illustrated in Fig. 2, consists of a stainless steel cylinder to contain the solution, covered with a stainless steel lid to which is attached an Aminco stainless steel heater element, the heating part of which is bent into a flat coil in such a manner that it may be lifted out with the lid. The temperature is regulated by a bimetal regulator in a stainless steel sheath inserted through a hole near the bottom of the cylinder and welded in place. The lid is cut out to allow access to the fluid but the cut out part is covered by a hinged cover whenever the fluid is not in continuous use. The whole apparatus, as far as the electric couplings, but without the bimetal thermoregulator, may be autoclaved. The thermoregulator is inserted and the couplings joined just before use. The 110 volt a.c. current in the heating element is controlled by an Aminco d.c. mercury relay. The current for the relay is supplied at 6 volt d.c. by an Aminco rectran connected to the thermoregulator.

Both of these methods are being used on a trial basis in the operating rooms of this Institute, the second apparatus being preferred. Work is in progress to simplify the second apparatus by designing a thermoregulator which may be sterilized and immersed directly in the solution.

A third apparatus for the same purpose consists of a coil of stainless steel tubing immersed in a constant temperature water bath. One end of the coil is connected by

* This apparatus was designed and built with the able assistance of Mr. L. Geddes.

rubber tubing to a glass tube dipping into the vessel containing the solution, the other end is connected by rubber tubing to a double-valve irrigation syringe.[1] The glass, steel, and rubber tubing may all be sterilized. By working the syringe, fluid is drawn through the coil and thereby warmed as it is used. The difficulty in obtaining

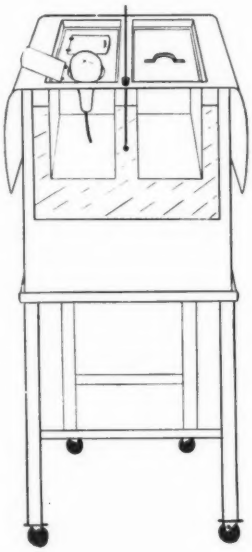


FIG. 1. Tank for holding sterile fluid in constant temperature bath.

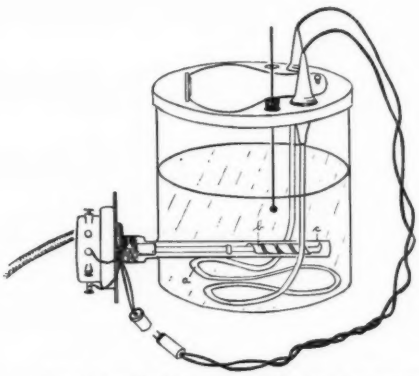


FIG. 2. Sterilizable apparatus for keeping sterile fluid at constant temperature. (a) Stainless steel heating element. (b) Bimetal thermoregulator. (c) Stainless steel sheath for thermoregulator.

syringes satisfactory for the purpose, and the cooling which occurs in the tube, render this method impractical for most purposes.

EXPERIMENTAL METHODS

In order to study the effects of irrigation fluids of different composition applied to the pia-arachnoid surface of the cortex under reasonably controlled conditions, a special irrigation chamber was constructed. This was a modification of the Forbes window[8] which has been used for microscopic observations of pial vessels in the living animal. It consists of a dural cylinder of 13 mm. inside diameter threaded at the base so that it could be screwed tightly into a trephine opening in the skull. A disc of thin optical glass was cemented into the top in some experiments so that microscopic observations and photographs could be made of the cortical surface with dura removed. Around the rim of the top of the cylinder there were several openings to provide for continuous perfusion of fluid through the chamber, for measurements of pressure within the chamber, the insertion of glass pH electrodes, a thermocouple for local temperature measurements within the chamber, and for the insertion of electrodes for recording the electrical activity of the cortex and for its electrical stimulation. This chamber is illustrated in Fig. 3. It has proven very useful in a number of experiments in which various

aspects of the reaction of the cortex to changes in surface chemical environment were to be studied.

The temperature of the exposed brain surface was found to be about 34°C. The irrigation fluid was admitted at about this temperature. This was achieved either by keeping the reservoir in a thermoregulated bath or by passing the fluid through coils in such a bath.

Measurements of the pH of the brain surface and of the irrigation fluid were made by means of micro glass electrodes and a silver-silver chloride-saline reference electrode similar to those described by Nims.[14] The elec-

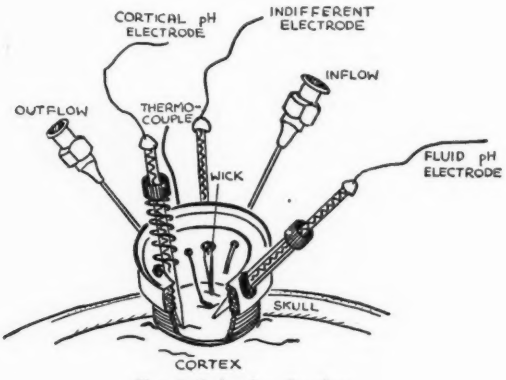


FIG. 3. Irrigation chamber.

trodes were connected to a Beckman pH meter through a switch box which allowed readings from 3 glass electrodes to be taken in rapid sequence. The wires from the glass electrodes, and the switch box, were shielded. It was necessary to ground the shielding and the water bath, while the operating table had to be carefully insulated, in order to obtain reliable and stable readings. Each glass electrode was calibrated against buffer solutions of known pH. The reference electrode was placed, in earlier experiments, against the cleaned surface of the skull of the animal; later the wick was immersed in the fluid in the irrigation chamber. The glass electrode was supported by a glass tube or light spiral wire sealed in the irrigation chamber. It was placed firmly, but not too heavily, against the cortex in the irrigation chamber to record the pH of the brain surface, or it was raised slightly in order to record the pH of the irrigation fluid. The pH of blood was determined on samples, drawn under oil, by means of a Beckman "hypodermic glass electrode" at room temperature. The value obtained was corrected for temperature according to Rosenthal[17] to obtain the value at 38°.

Experiments were carried out on cats under light nembutal or dial-urethane anaesthesia. In some experiments, artificial respiration was maintained.

pH EFFECTS

The pH of the Irrigation Fluid. Unbuffered solutions, such as Solution A or normal saline, usually show a pH of about 6 due to dissolved CO_2. When the flow of unbuffered solution through the irrigation chamber was slowed down, the pH of the fluid usually fell further, sometimes as low as 5.5. This low pH presumably resulted from more rapid diffusion of CO_2 than of base from the brain surface into the fluid. When the flow was stopped, leaving fluid static in the chamber, the pH gradually rose, reaching 6.3 to 6.8 in about 5 minutes. The pH of these solutions was never observed to reach that of the brain surface though theoretically that might be expected ultimately.

Even an appreciably alkaline, but unbuffered, salt solution (0.002 per cent added sodium carbonate, pH over 8) acquired a low pH when flowing slowly over the brain. The carbon dioxide diffusing from the brain will presumably reduce any solution to a relatively low pH unless the solution contains sufficient buffer to take care of the approximately 5 per cent carbon dioxide tension which is present at any tissue surface. For instance, a saline solution containing 0.01 M phosphate buffer at pH 7.5 changes to about pH 6.7 in the presence of 5 per cent carbon dioxide at 37°. But even with a phosphate concentration as low as 0.01 M, the phosphate causes the calcium of a balanced salt solution to precipitate on standing or warming the solution. Bicarbonate, which is the physiological buffer, is the only buffer salt that may be introduced in sufficient concentration to control the pH without rendering the fluid highly unphysiological.

A bicarbonate-containing medium, such as Solution B or spinal fluid, in equilibrium with 5 per cent carbon dioxide at 38° has a pH of about 7.4. But this equilibrium is maintained only in a closed system, or if 5 per cent carbon dioxide is bubbled vigorously through the solution. Such a solution on exposure to air loses carbon dioxide and alkalinity develops. When the irrigation chamber was filled with long exposed Solution B, with a pH of 8.0 to 8.8, and the flow stopped, the pH of the fluid fell rapidly until a pH of about 7.4 was reached, and then remained constant. The rate at which this change occurred naturally depended upon the distance through which the carbon dioxide escaping from the brain surface had to diffuse into the static fluid. When the glass electrode was very close to the brain surface, the new pH was stabilized at about pH 7.4 within about 30 seconds. Records illustrating these various effects are shown in Fig. 4.

Thus a properly constituted bicarbonate solution may have a pH slightly different from that of normal blood or spinal fluid, usually more alkaline, but, in contact with the brain surface, it rapidly assumes approximately the same pH as the blood. But solutions lacking bicarbonate are usually initially slightly acid, tend at first to become still more acid, and do not achieve the physiological pH within a reasonable period.

Reactions of Pial Vessels. The pial vessels were found to be very sensitive to the pH of the irrigating fluid. Whenever a fluid of pH below 7 was in

the chamber, the pial vessels were markedly dilated, the whole surface appeared flushed, a network of previously invisible fine vessels became apparent, and sometimes petechial hemorrhages appeared. But when such a fluid was replaced by the bicarbonate-buffered fluid, constriction of the vessels and a blanching of the whole brain surface occurred. These changes

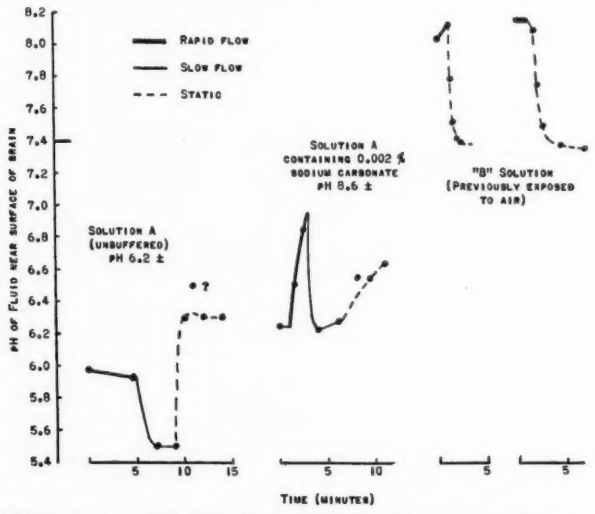
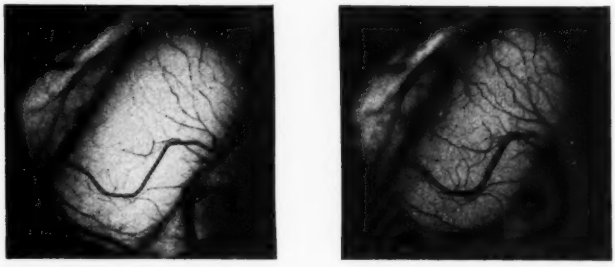

RAPID FLOW
SLOW FLOW
STATIC
SOLUTION A (UNBUFFERED) PH 6.2 ±
SOLUTION A CONTAINING 0.002 % SODIUM CARBONATE PH 8.6 ±
"B" SOLUTION (PREVIOUSLY EXPOSED TO AIR)
PH OF FLUID NEAR SURFACE OF BRAIN
TIME (MINUTES)

FIG. 4. Effect of contact with the brain surface on the pH of various solutions.

are illustrated in the photomicrographs of Fig. 5. The extent of the vessel dilatation or constriction produced by different fluids seemed to be correlated with the pH recorded from the fluid in the chamber. Even Ringer solution containing a trace of sodium carbonate, which, as shown above, acquired a low pH in contact with the brain, produced hyperemia. Solution B



a b

FIG. 5. Photomicrographs of the pial vessels of a cat brain. (a) under Solution A (unbuffered), (b), under Solution B (bicarbonate-buffered).

produced the same, presumably normal, blood-vessel picture as human spinal fluid when either of these had been static in the chamber long enough for pH equilibrium to be reached. The reaction of the vessels to change of irrigation fluid was not instantaneous. The vessel constriction or dilatation which occurred on changing from unbuffered irrigating solution (low pH) to bicarbonate-containing solution (physiological or higher pH) or vice versa, usually took up to 30 seconds or more to become obvious.

After long exposure of the brain the responsiveness of the vessels to change in irrigation fluid often diminished or disappeared. In these cases the vessels seemed to be in an irreversibly dilated condition; irregular dilatation or beading of vessels was often noted. We have not been able to relate this loss of responsiveness and apparent loss of vessel tone to the nature of the irrigation fluid. But we feel that dilatation and constriction of the pial vessels in response to changes in the irrigating fluid provide a useful test for the condition of the pial vessels which should help in further elucidation of factors that affect the exposed brain.

It should be pointed out that the dilatation produced by unbuffered solution is almost certainly due to low pH and not to a chemical effect of the dissolved carbon dioxide which produces the low pH. The bicarbonate-buffered medium, which produces the opposite effect, has a higher pH but at least an equal concentration of dissolved carbon dioxide and a much higher concentration of bicarbonate ion.

Observations similar to those made on cats have been made on a dog and on human subjects (with Dr. W. Penfield) during operations involving craniotomy. When the exposed brain was irrigated with Ringer solution the surface of the brain became flushed. But on turning back a flap of dura, which had protected the underlying surface from the irrigation fluid, this surface was observed to be still pale. Further, the flushing took much longer to develop under an area of thickened arachnoid. Areas of the brain lying under collections of spinal fluid remained pale and irrigation with Solution B produced in some instances a paler brain than is observed with unbuffered irrigation fluid. However, as in the animal experiments, this was not always true. Other factors may cause hyperemia during an operative procedure, besides the low pH of the usual irrigation solutions.

The pH of the Brain Surface. The pH of a given point on the brain surface varies with changes in blood pH as influenced by the respiration (Dusser de Barenne *et al.*[5,6]) It is raised briefly when the local circulation is increased as a result of increased neuronal activity and then falls (Jasper and Erickson[11]). The rise is presumably due to the pH of the blood being somewhat higher than that of the brain-tissue fluids. (However, none of the above authors gave values for the actual pH of the blood.) The fall in pH is presumably due to excessive production of acid metabolites during neuronal hyperactivity.

We have observed, on cats and monkeys, that the pH recorded from different points on the cortex varies appreciably. Values between 7.07 and 7.34 have been obtained during a short period when there was no obvious

variation in the condition of the animal. Values of 7.0 and slightly lower have been recorded from cat, dog, and monkey cortex. At a given point, under steady conditions, the pH remains constant or shows only a slow drift. The highest values for the cortex were equal to or slightly below that

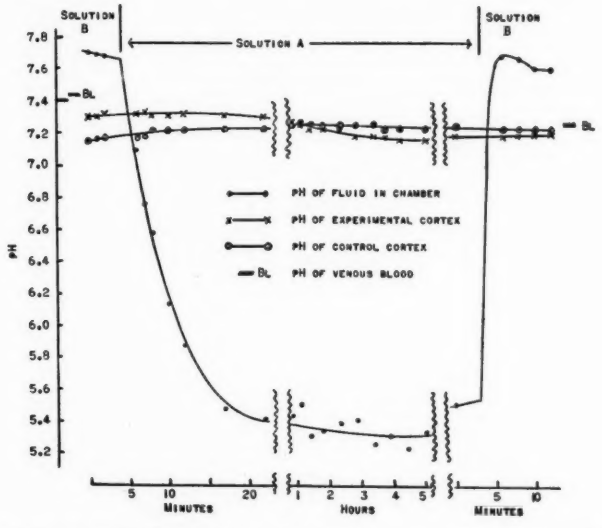


FIG. 6. Effect of pH of the irrigating fluid on the pH of the cortex. The glass electrode recording the pH of the experimental cortex was placed against the brain surface in the perfusion chamber. The electrode recording the pH of the fluid was in the same chamber but its tip was about 1 mm. above the brain. The control cortex electrode was placed against the brain surface in another chamber on the opposite side. The control chamber was filled with Solution B nearly an hour before the record was started and this solution remained there throughout so that it was presumably in equilibrium with the tissue fluids.

of the venous blood at 38° (with one dubious exception). The pH of arterial blood is about 0.05 higher than that of venous blood, and at the temperature of the exposed cortex, 34° ±, either blood pH would perhaps be about 0.07 higher.[17] The pH of the cortex surface thus tends to be appreciably lower than that of the blood supplying it.*

The variability from point to point was apparently connected with the proximity of blood vessels. Higher values were in general obtained when the electrode was on or near a large vessel. However the diameter of the electrode tips used was somewhat more than 1 mm. and the use of finer electrodes will be necessary for more precise mapping of the pH of the cortex in relation to blood vessels. A similar but more precisely observed and much more marked variation in the oxygen tension of the cortex in relation to the proximity of blood vessels has been observed by Bronk *et al.*[3]

* It was interesting to observe that, on asphyxiating the animal by constriction of the trachea, the pH of the cortex fell rapidly, reaching 5.9 in 20 minutes. Blood drawn from the heart of the animal killed in this manner had a pH of 6.8. The rapid fall in pH of the brain is presumably mainly due to anaerobic glycolysis (lactic acid production) at the expense of glucose remaining in the brain.

The pH of the brain tissue, as indicated from an electrode placed on the surface of the cortex, was not appreciably affected by wide variations in the pH of the irrigating fluid. This observation is illustrated in the experiment recorded in Fig. 6, in which it is shown that even long contact with a fluid of low pH has no obvious effect on the pH of the cortex. Similar observations were made a great many times on numerous cats. This maintenance of pH is probably aided by the changes in pial circulation mentioned above though similar constancy of pH has been noted at a time when the reactivity of the pial vessels had been apparently largely lost. Presumably the main control rests in the high buffer capacity of the brain fluids and it seems as if there is some kind of barrier to the free inter-diffusion, between the tissue fluids and the irrigating fluid, of factors that affect pH. (It is perhaps relevant here to note that some time, up to 10 minutes, is often required for an electrode freshly placed on the cortex to give a steady reading. Presumably this time is required for the fluid in the concavity of the electrode tip to reach equilibrium with the brain fluids. Once placed and allowed to reach equilibrium, the electrode records changes in local pH within a few seconds.[11])

DISCUSSION

No obvious difference was observed in the immediate vascular reactions produced by Solution A, other Ringer-type solutions or normal saline. (A strongly hypertonic solution, 1.8 per cent sodium chloride, produced a much more pronounced reaction to the extent of petechial hemorrhages.) The marked dilatation of the superficial vascular bed of the cerebral cortex in response to the isotonic salt solutions now in current use seems to imply an irritating action of these unbuffered solutions which may play a role in the enhancement of damage caused by prolonged exposure of the cortex during neurosurgical procedures. The relative importance of the composition of the irrigation solutions in relation to other aspects of the abnormal environment in which the exposed cortex finds itself has yet to be ascertained. It is possible that control of the pH, by the use of bicarbonate, is only one of several ways in which exposure damage may be minimized through improvement in the irrigation fluid. The addition of glucose to the medium has strong support in principle but as yet its value has not been demonstrated. The presence of protein or other material exerting colloid osmotic pressure in the irrigation fluid might have value. A solution, using gelatin as the colloid, has been prepared but has received only preliminary trial. Prados, Strowger and Feindel[16] reported that administration of adrenal cortical extract reduces the delayed vascular reaction to exposure of the cortex in cats. These authors noted that the extract was effective even when applied to the exposed brain. The possibilities of incorporating such a hormone in the irrigation fluid are therefore also being considered.

SUMMARY

1. Unbuffered isotonic salt solutions in contact with the brain surface cause marked dilatation of the pial blood vessels. This hyperemia may be

prevented by the use of bicarbonate buffered solutions at physiological pH or higher.

2. Unbuffered solutions in contact with the brain surface rapidly assume a relatively low pH. A bicarbonate buffered solution may be faintly alkaline initially but, in contact with the brain surface, it rapidly assumes the physiological pH.

3. The pH of the pia-arachnoid surface of the cortex remains essentially constant in spite of wide variations (5.3 to 8.0) in the pH of the fluid irrigating the surface in its immediate vicinity.

4. A perfusion chamber suitable for the study of the effects of various dissolved chemical agents on the pH, electrical activity and other physiological functions of the cerebral cortex is described.

5. Practical methods for the preparation and dispensation of two sterile solutions closely resembling spinal fluid and suitable for irrigation of the brain or other tissues, and for replacement of spinal fluid are described. One of these is a simply modified Ringer solution, the other contains bicarbonate in physiological concentration.

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PARAPHYSIAL CYSTS OF THE THIRD VENTRICLE
WITH REPORT OF EIGHT CASES*

JAMES GREENWOOD, JR., M.D.
Methodist Hospital, St. Joseph's Infirmary, and Baylor University College of Medicine, Houston, Texas

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THE PARAPHYSIAL cyst is one of the most favorable brain tumors with respect to operative mortality. Despite the fact that it lies within the 3rd ventricle, a most vulnerable area of the brain, its fragile attachments make its removal relatively easy. There is a challenge to the surgeon in that removal must be accomplished without damage to the walls of the 3rd ventricle, for it seems that if this occurs permanent character and personality changes will result.

The gross appearance of the cyst (Fig. 4) is characteristic: a bluish or greyish appearing capsule with a gummy or rubbery feel. Nearly always there is a firm attachment to the choroid plexus at the anterior part of the roof of the 3rd ventricle (Fig. 7). The contents of the cyst are a thick, greyish or brownish colloid material and when looking into the lateral ventricle, the tumor nearly always can be seen presenting at the foramen of Monro. Microscopically there is a fibrous cyst wall with a lining of columnar epithelium (Fig. 6). Goblet cells and cilia have been described.

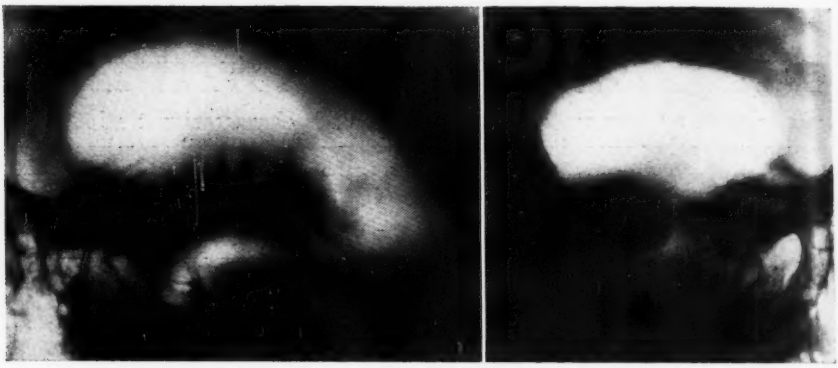
Bailey[1] as early as 1916 studied the paraphysis in human embryos and the possibility of a relationship of this structure to these tumors. There were many isolated reports in the literature, but there were no successful surgical attempts until Dandy[2] in 1933 reviewed 31 autopsies in the literature and reported 5 operated cases of his own, all but 1 of his patients having survived surgery. He doubtless encountered others later, but did not report them. Zimmerman and German[16] in the same year reported 2 cases with 1 death. Stookey[9] in 1934 placed emphasis on the intermittent symptoms. Rehbock[6] in 1936, in reviewing 56 cases, including 3 of his own, noted that in 14 per cent death occurred in the initial attack. McLean,[5] Trescher and Ford,[11] Gardner and Turner,[3] Zeitlin and Lichtenstein,[14,15] Traut and Piette,[10] Larson,[4] Weinberger and Boshes,[12] Shannon,[7] and Wilson[13] all reported 1 or 2 cases. Shaver[8] reported 4 cases, 2 of the patients having been operated upon. A total of something over 60 cases has been reported, and in 15 of these operations were successfully performed. There are, of course, many unreported successful surgical cases.

I wish to report 5 operated cases of my own, and 3 additional autopsy cases, included through the courtesy of Baylor University and the Houston City Health physician. These last 3 are of particular importance, since they greatly emphasize the need for emergency ventriculography in cases of sudden coma where there is no evidence of infection, diabetes, or nephritis.

* Read before the Central Neuropsychiatric Association, October 1947 (one additional case since).

In reviewing the literature, the three most popular names for this tumor are colloid cysts, neuroepithelial cysts, and paraphysial cysts. The last seems most desirable. If the paraphysial origin is not correct, one would expect to find the tumor arising in other ventricles, or at least at other points in the 3rd ventricle. The name is good from the standpoint of position and should be used until a better one is found.

The neurological symptoms are very meager and seldom are sufficient to make a diagnosis. A patient may have intermittent headaches for years, but once the foramen of Monro is blocked the course of events proceeds rapidly. Blocking may be sudden, since the tumor may act as a ball valve. Incontinence of urine, nystagmus, parkinsonian rigidity, and other signs of little localizing value are seen occasionally. Choked disc occurs in approximately half of the cases.



FIGS. 1 and 2. Paraphysial cyst. *Left*, Lateral ventriculogram. No air shown in 3rd ventricle except for sickle-shaped filling below tumor. *Right*, Good filling of anterior part of 3rd ventricle achieved by lateral view with the face up and head hyperextended.

A peculiar type of organic mental reaction has occurred in all of the 5 operated cases. This consists of varying degrees of disorientation, particularly for time and place, while the patient superficially appears to be alert and mentally normal. Only careful checking has revealed this reaction. It is similar to Korsakoff's psychosis in that the patient fills in his memory defects with confabulation. In all cases, this feature cleared up within 2 to 3 months after operation.

RADIOLOGY AND SURGERY

During the ventriculography, usually it can be demonstrated that there is no free communication between the lateral ventricles. The ventricles will contain a large amount of fluid, and plates taken following replacement of the fluid with oxygen will show dilated lateral ventricles and often a deviated septum pellucidum (Fig. 3). With the patient on his back and the head hy-

perextended, a lateral view (Figs. 1 and 2) will show the anterior part of the 3rd ventricle in the shape of a sickle, thus outlining the anterior border of the tumor.

Surgery is not difficult. Three methods of approach have been used: (1) transcortical, (2) through the posterior corpus callosum, and (3) through the anterior corpus callosum. Of these, the transcortical is somewhat the easiest, but has the disadvantage of all cortical resections in that postoperative convulsions may result. This is less likely if a block of the frontal lobe

FIG. 3 (*left*). Typical anterior view showing absence of gas in the 3rd ventricle and deviation of the septum pellucidum.

FIG. 4 (*right*). Three of the five paraphysial cysts removed surgically.

is resected than if a simple incision is used to enter the ventricle. Approach through the posterior corpus callosum does not produce as good an exposure, but has no other disadvantages. Anterior callosal section produces ideal exposure and minimal possibility of convulsions, but care must be used in protecting the anterior cerebral artery, which should be covered carefully with moist cotton. Once the lateral ventricle is entered, the foramen of Monro may be widened by incisions, both anteriorly and posteriorly, to allow delivery of the cyst. Several vessels usually need to be divided and part of the choroid plexus is removed with the tumor. Care must be taken not to damage the walls of the 3rd ventricle.

OPERATIVE RESULTS

There was 1 death in the 5 cases in which operation was performed, and 2 of the 4 survivors are working. Case 1 (Table 1), who was 59 years of age at the time of operation, was able to do some work for over a year, but at the age of 63 showed considerable mental deterioration and died in June 1947, 7 years after surgery. All 4 patients had one or more convulsions after operation, and 2 had convulsions after the first postoperative year. Case 4

did fairly well for about a year, but developed a psychoneurosis. A report from a veterans' hospital stated that there was no mental deterioration.

ILLUSTRATIVE CASES

Case 3. R. D., a white male aged 14 years, was first seen in the Methodist Hospital Aug. 21, 1942, because of severe headache and vomiting of sudden onset 18 days before. Except for questionable haziness of the disc margins, the neurological examination was negative. All of his symptoms cleared up following spinal puncture,

TABLE 1

Case summaries: Paraphysial (colloid) cysts of the third ventricle

Case	#1 A.E.O.	#2 C.C.D.	#3 R.D.	#4 S.J.M.	#5 E.M.M.	#6 J.D.H.	#7 S.K.	#8 I.E.
Sex	M	M	M	M	F	M	M	F
Age	59	37	14	50	39	8	26	25
Duration Symptoms	4 yrs.	1 mo.	3 mos.	3 wks.	8 mos.	1 day	2 yrs.	1 day
Intermittent Attacks	+	+	+	0	+	0	+	0
Headache	+	+	+	+	+	0	+	+
Vomiting	0	+	+	0	+	0	+	0
Convulsions	0	0	0	0	0	+	0	+
Unconscious Attacks	+	+	0	+	+	+	+	+
Papilledema	1D	0	2D	4D	3D	0	0	0
Disorientation	+	+	0	+	0	0	0	0
Parkinsonism	0	0	0	+	0	0	0	0
Babinski	Rt.	0	0	0	0	0	0	0
Strabismus	0	+	0	0	0	0	0	0
Nuchal Rigidity (mild)	0	0	0	0	0	0	+	0
Spinal Fluid Pressure	400	not done	normal	not done	not done	not done	not recorded	not done
X-ray	erosion sella	neg.	neg.	neg.	erosion sella	"	"	"
Hydrocephalus	marked	marked	marked	moderate	marked	"	"	"
Operative Approach	trans-cortical	cortical resection	cortical resection	trans-callosal	trans-callosal	"	"	"
Size of Tumor	2 cm.	1.5 cm.	2 cm.	2 cm.	2 cm.	1 cm.	2.5 cm.	1.2 cm.
Recovery	yes	yes	yes	yes	no	All Died Within 24 Hrs.		
Ability to Work	60%	80%	95%	75%	0	"	"	"
Present Status	died 7 yrs. postop.	good 6 yrs.	good 6 yrs.	poor 5 yrs.	0	"	"	"
Necropsy	no	living	living	living	clot in aqueduct	+	+	+

In the case summaries attention is called to the fact that 2 patients had no history of a previous attack and died within 24 hours. Ventriculography was necessary for diagnosis in all 5 operated cases. Localizing neurological symptoms were notably absent. One case showed a Babinski sign, 1 strabismus, 1 definite parkinsonism, and 1 paresthesia in one arm. General symptoms of sudden increased pressure were prominent. All but 1 had severe headache and unconscious attacks. Four had papilledema.

which showed a normal pressure. Three weeks later, an encephalogram was done because of recurrence of symptoms. The ventricles were poorly filled and again his symptoms cleared up. Six weeks later his headache returned, and this time he had

definite papilledema. A ventriculogram showed large ventricles which did not communicate, and there was no gas in the 3rd ventricle. Through a right frontal craniotomy, a small cortical resection was done and a bulging septum pellucidum was punctured. A bluish cystic tumor was visible at the foramen of Monro, which was further exposed by enlargement of the foramen. The cyst was then partially aspirated and removed.

His convalescence was uneventful except for mild disorientation which lasted

FIG. 5. Autopsy specimen of one of the coroner's cases (Case No. 7). Corpus callosum and tela choroidea removed.

about 4 weeks. He made good grades in school, pitched on the baseball team, carried a paper route, and eventually obtained permanent employment with an oil company. He had three convulsions following surgery and has had two slight attacks since, but none in the past 2 years.

Pathological report: The cyst was $1\frac{1}{2} \times 2$ cm., with a thin wall, containing dark, thick brownish fluid. In the wall was a calcified mass 0.5 cm. in diameter. Microscopic examination showed a fibrous capsule lined with a single layer of epithelium.

Case 7. S. K., a 26-year-old white man, was admitted to the Jefferson Davis

Hospital Mar. 11, 1947, acutely ill with severe headache. He had been to his family doctor that morning because of headache and vomiting, and had fainted in his office. He had had several such attacks during the past 2 years. On admission, the right pupil was dilated and there was a voluntary opisthotonos, but no real nuchal rigidity. Spinal puncture revealed no increase in cells; the pressure was not recorded. Death occurred the same evening from respiratory failure.

Necropsy showed an edematous brain with dilated ventricular system. In the anterior part of the brain was an elongated, thin-walled cyst (Fig. 5), 2×2.5 cm., containing yellow, cloudy fluid. It was attached to the choroid plexus.

FIG. 6 (*left*). Photomicrograph. Note layer of columnar epithelium and cilia.
FIG. 7 (*right*). Portion of cyst wall, showing attachment to choroid plexus (upper right).

COMMENT

In a series of over 300 brain tumors, 5 were paraphysial cysts of the 3rd ventricle. This is a rather high percentage for this supposedly rare tumor. Three additional cases, supplied by the coroner and the pathology laboratory, emphasize the dangerous position of these tumors and illustrate why some of them never reach the neurosurgeon.

SUMMARY AND CONCLUSIONS

1. The etiology, pathology, symptoms, and diagnosis of paraphysial (colloid) cysts of the 3rd ventricle have been discussed.

2. Five operated cases have been reviewed and 3 additional cases solved by postmortem examination have been included.

3. The diagnosis of paraphysial cyst can be proved only by ventriculography, but an intermittent, severe organic headache should arouse suspicion.

4. The term "paraphysial cyst" seems at the present time to be the most accurate and descriptive name for this tumor.

5. Improvement in ventriculography by a lateral view taken with the head well back in hyperextension is suggested.

6. Approaching these tumors through the anterior corpus callosum after separation and protection of the two anterior cerebral arteries is believed to be feasible.

7. The need of emergency ventriculography in cases of rapid, unexplained coma is emphasized, particularly if there has been a previous similar attack.

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RESULTS OF OPERATIONS FOR LUMBAR PROTRUDED INTERVERTEBRAL DISC*

JOHN RAAF, M.D.,† AND GEORGE BERGLUND, M.D.
Portland, Oregon

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While there is now agreement on most points in diagnosis and treatment of lumbar protruded intervertebral disc, some questions regarding the subject are still controversial. Should myelography be employed as a diagnostic aid or should the surgeon depend entirely upon symptoms and signs? What are the results of the surgical treatment of protruded intervertebral disc? Is the patient able to return to his former employment? What is the average time-loss of a workman from his job, and when compensation cases are closed, what is the average permanent partial disability award? How much does the average case cost the insurance carrier? What factors may influence the end results? Is a better result obtained when a protruded disc is found and removed than when there is a negative exploration or when exploration reveals an inflamed nerve root, adhesions about the nerve root, or some other pathological condition? Do private patients report better results than compensation patients? How often should spinal fusion accompany the removal of the protruded portion of the intervertebral disc? Often one has an impression, which may be erroneous, because of the impact one or two outstanding cases has made on his memory. For this reason, an evaluation of a series of consecutive cases was undertaken, and the questions enumerated constitute the problems discussed in this paper.

CASES STUDIED

During the ten-year period from 1938 to 1947, inclusive, 224 patients were operated upon by one of us (JR) for protruded intervertebral disc in the lumbar area. No thoracic or cervical protruded disc cases were included in this study. Detailed preoperative examinations were carried out in the office or in the hospital by one or more members of our staff, and all operations were performed in private hospitals. No indigent patient operated upon in a charity hospital was included in the series. Of the 224 cases, 129 were compensation cases. Classed as "compensation cases" are those patients who not only had their medical bills paid by an insurance carrier but who also drew compensation while they were off work and who were given a permanent disability award if any disability remained at the time their cases were considered stationary and, therefore, were closed. The compensation cases were covered by a variety of insurance carriers—11, in all. Ninety-five of the 224 cases were "private."

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† 912 Medical-Dental Building, Portland 5, Oregon.

THE VALUE OF MYELOGRAPHY

We have used lipiodol, air, thorotrast and pantopaque as contrast media in the myelographic studies. Table I shows an analysis of the results with lipiodol, thorotrast and pantopaque. None of the 6 patients who had air myelograms came to surgery, so this type of myelography has not been evaluated. If the contrast media indicated the presence of a protruded disc at a certain level, yet at surgery the protrusion was not found there, or if, in spite of a negative myelogram, surgery was undertaken and a protrusion found, an error for myelography was recorded. Twenty-seven lipiodol studies were performed and the method was 88.9 per cent accurate in locating the presence and the level of the protrusion. The 4 thorotrast studies were 100 per cent accurate and the pantopaque studies 76.7 per cent accurate. Under the following two conditions an error was recorded for diagnosis based on

TABLE 1

Accuracy of diagnosis based on contrast media and on clinical signs and symptoms

Contrast Media	Number of Studies	Myelography Proven Correct At Surgery	Myelogram Indicated Disc But No Disc Found	Myelogram Indicated No Disc But Disc Found	Total Errors
Lipiodol	27	24 (88.9%)	3	0	3 (11.1%)
Thorotrast	4	4 (100%)	0	0	0
Pantopaque	103	79 (76.7%)	21	3	24 (23.3%)

Diagnosis Based On Clinical Signs and Symptoms	Clinical Opinion Proven Correct	Disc Thought To Be Present But No Disc Found	Operation Would Not Have Been Done on Clinical Signs—Myelography Positive—Disc Found At Operation	Total Errors
224	182 (81.25%)	30	12	42 (18.75%)

clinical signs and symptoms: if the disc was thought to be present but no protrusion was found at operation, or if the symptoms and signs were not sufficiently indicative of a protruded disc to justify surgery but a protrusion was found and removed on the strength of the myelographic study. Our accuracy of diagnosis from symptoms and signs alone was found to be 81.25 per cent.

When a pantopaque test is skillfully performed, there is little or no discomfort to the patient and the oil can be removed completely in practically all instances. Without pantopaque studies some of the multiple protrusions will be missed. Then, too, there is no clinical sign that will invariably designate the exact site of the protrusion. We believe a pantopaque study performed to definitely locate the protrusion is less harmful to the patient than exploration of extra disc spaces. The more one limits surgery and the less one disturbs articular facets, the better the end result.

While we realize that myelography, like any other laboratory test, is not infallible, we see no reason to change our current policy of carrying out a pantopaque study in a high percentage of cases. In the last 2 years, 71 per cent of the patients whom we explored for protruded intervertebral disc had a preoperative pantopaque study. If the pantopaque study revealed no protruded disc and yet the clinical signs were sufficiently indicative of a protruded disc, the myelogram was disregarded. Pantopaque studies are at the present time not usually done upon patients when the diagnosis, as well as the level, seems very definite.

POSTOPERATIVE RESULTS

Fairly wide laminectomies were done on some of the first patients in the series while all those operated upon in recent years had interlaminal explorations with little or no bone removed except when a very large protrusion was present.

The operative mortality for the 224 patients was zero. Since operation, 2 patients have died of causes entirely unrelated to the protruded intervertebral disc. Acute, bacterial endocarditis was the cause of one death $1\frac{1}{2}$ years after operation, and cerebral hemorrhage was responsible for the other death $2\frac{1}{2}$ years after operation.

Five of the 224 patients required second operations. Re-exploration showed 2 of the 5 had recurrences of the protrusions, while in 3 no recurrence was found. Following re-exploration and spinal fusion, the 2 with the recurrences and 1 other have been relieved, while 2 of the 5 are no better.

Of the total 224 patients the end results following surgery were evaluated upon 160. The others were eliminated from the follow-up study for one or both of the following reasons: 1) sufficient information was unavailable regarding the patient's present condition, and 2) less than 1 year has elapsed following operation. Information regarding the results came chiefly from three sources: 1) office and hospital records; 2) questionnaires answered by the patients; 3) records of the insurance carriers in those cases covered by compensation.

A questionnaire was sent to all of the patients who had been explored for protruded intervertebral disc during the years 1938 through 1946, inclusive, and was answered by 117. One of the questions asked was whether the patient thought he was completely cured, 75 per cent better, 50 per cent better, 25 per cent better, the same as before operation, or worse than before operation. Chart I gives the results of the answers.

The questionnaire also asked whether the patient had been able to go back to the same type of work or work equally as arduous as he had been doing prior to the onset of symptoms, whether he had returned to work but was doing lighter work, or whether he was unable to work (Chart II). Of the 117 patients, 95 (81.2 per cent) had returned to the same work, 10 (8.55 per cent) had returned to lighter work, 9 (7.69 per cent) were unable to work be-

Chart I

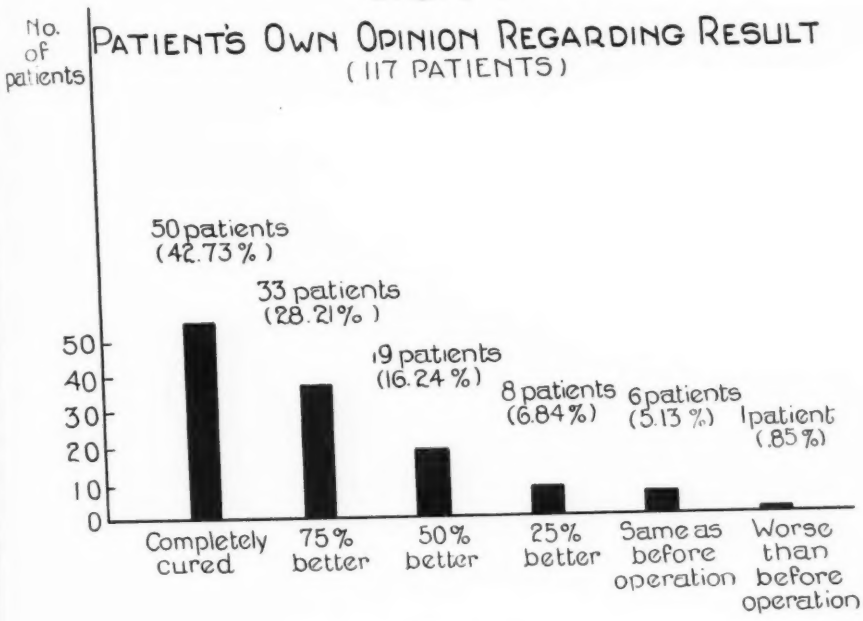
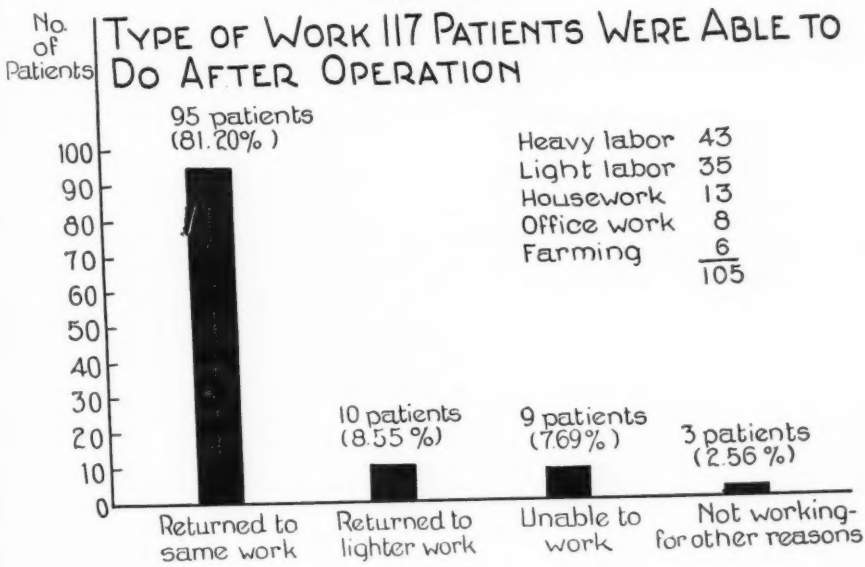


Chart II



cause of symptoms, and 3 (2.56 per cent) were not working because of other reasons. The type of work being performed by the 105 patients who are working after operation is indicated in the insert on Chart II. There are 43 performing heavy labor, 35 light labor, 13 housework, 8 office work, while 6 are farming.

We realized that the questionnaires returned by the 117 patients might not represent a true picture of the results, so we sought further information by obtaining permission to examine the records of the insurance carriers in the compensation cases. It was possible to examine the records of 88 compensation patients who had been operated upon 1 to 10 years previously. Table 2 lists some of the information obtained from the insurance carrier records.

The total number of working days lost is figured to the day the insured's claim is closed. The patient might not start back to work as soon as his claim is closed, but the claim is not closed until the case has become stationary and it is deemed by medical examiners that the claimant (1) is permanently totally disabled; (2) is partially disabled but can return to some type of work; or (3) has recovered completely from the symptoms caused by the protruded intervertebral disc. In the latter two instances, therefore, the patient is ready for some type of work. The average cost ($3,188.75) to the insurance carrier includes (1) the medical costs in the form of hospital, drug, transportation, nurse, and doctor bills, (2) compensation paid while the patient is being treated, and (3) final compensation award. Three patients were considered unable to do any type of work again and, therefore, were awarded permanent total disability; 2 of these had had their backs operated upon once before our operation was performed. Eight patients were considered completely cured so far as their protruded discs were concerned, and 77 were given some degree of permanent partial disability. The average permanent partial disability award was $1,015.40

TABLE 2

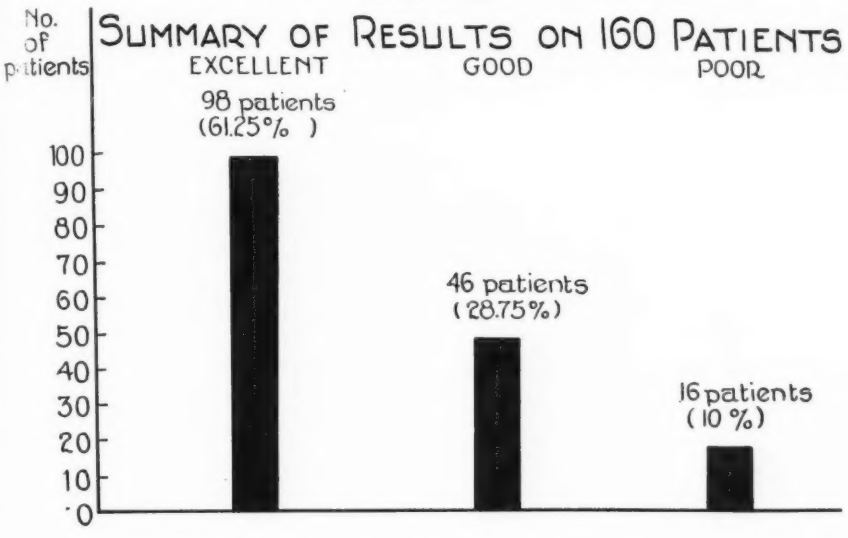
An analysis of 88 compensation cases

Closed with permanent partial disability award	77
Closed with no award	8
Closed with permanent total disability award	3
Average working days lost	356.5
Average cost to insurance carrier	$3,188.75
Average permanent partial award to worker	$1,015.40

When all available evidence was gathered from the 117 questionnaires, the hospital and office records, and the insurance carriers, we believed we had enough information on 160 patients who had been operated upon 1 to 10 years previously to be able to classify the result of the operation we had performed as excellent, good, or poor. An excellent result means that the patient is satisfied, that he is back at his former occupation or performing some duty equally as arduous as the work he had done before the onset of

symptoms, and that he is having little or no discomfort. A good result means that the patient is working but is not performing as heavy labor as he had done before the onset of symptoms, and that he is having some pain and might, at times, have to stop work to give his back a rest. A case was classed as a poor result if the patient is able to do little or no work and he feels that he is no better or is worse than before operation. This study of 160 patients revealed that 98 (61.25 per cent) had excellent results, 46 (28.75 per cent) had good results, and sixteen (10 per cent) had poor results (Chart III).

Chart III



FACTORS INVOLVED IN THE RESULTS

An attempt was made to determine whether there were any factors that might influence the result. Using the 160 patients listed on Chart III, the condition found at operation was studied and correlated with the end result. Table 3 summarizes these results. As one would anticipate, an excellent or good result is much more likely to follow when a protruded intervertebral disc is found and removed than when there is a negative exploration or when an inflamed nerve root, adhesions about the root, or some other pathological condition is present.

It is the general impression that private patients report better results than compensation patients, and our own experience bears out this impression. Again using the 160 patients listed on Chart III, the results in the compensation and private patients are compared in Table 4.

One of the much debated questions is whether a spinal fusion should routinely accompany the removal of a protruded intervertebral disc. It is

TABLE 3

Correlation of results with pathological condition found at operation

Condition Found	Number of Patients	Results		
		Excellent	Good	Poor
Protruded intervertebral disc	133	92 (69.2%)	34 (25.6%)	7 (5.2%)
Inflamed nerve root	5	0	3 (60%)	2 (40%)
Adhesions about nerve root	5	2 (40%)	2 (40%)	1 (20%)
Large veins	5	2 (40%)	2 (40%)	1 (20%)
Osteoarthritic lipping	2	0	2 (100%)	0
Spondylolisthesis	1	0	1 (100%)	0
Congenital anomaly of L5	1	0	0	1 (100%)
No pathology found	8	3 (37.5%)	3 (37.5%)	2 (25%)
	160			

TABLE 4

Results in compensation and private patients

Type of Case	Number of Patients	Results		
		Excellent	Good	Poor
Compensation	108	57 (52.8%)	35 (32.4%)	16 (14.8%)
Private	52	40 (76.9%)	10 (19.2%)	2 (3.9%)
	160			

TABLE 5

Results in patients with and without spinal fusion

	Number of Patients	Results		
		Excellent	Good	Poor
Disc removed with fusion	40	26 (65%)	11 (27.5%)	3 (7.5%)
Disc removed—no fusion	93	65 (69.9%)	23 (24.7%)	5 (5.4%)
No disc found—fusion done	19	5 (26.3%)	9 (47.4%)	5 (26.3%)
No disc found—fusion not done	8		5 (62.5%)	3 (37.5%)
	160			

Compensation Cases

	Permanent Partial Disability Award	Days Lost From Work
Disc removed and fusion done	$1,168.86	381.1
Disc removed but no fusion done	872.77	315.2

obvious that it is impossible to arrive at an answer to this question from a statistical study, for rather than performing spinal fusions upon alternate patients in a series, we employ fusion where it seems indicated. Nevertheless, Table 5 shows a comparison of the results in the patients in whom a fusion was done with those in whom no fusion was done. The only conclusion to be drawn from these results is that it is not necessary to do a fusion in every patient from whom a protruded disc is removed in order to obtain a good result. Many of our patients are back at hard labor who have not had fusions. If the patient has had a great deal of back pain, if the roentgenogram shows a spondylolisthesis, a lateral defect in the neural arch, an inequality of the angle of articulation of the facets on the two sides, or any other evidence of instability, then a fusion should be done, especially if the patient is young and active. Also we are inclined to do a spinal fusion when at the time of surgery an instability of the 4th or 5th lumbar vertebra is discovered or when there is a negative exploration for protruded disc. We are of the opinion that the best results occur in those cases where there is close cooperation between the neurosurgeon and the orthopedist. Eighty-five (37.9 per cent) of the 224 patients in this series had spinal fusions at the same time the exploration for the protruded disc was carried out.

From a compensation standpoint a spinal fusion patient loses more days from work and has to be closed at a higher permanent partial disability rating than one who does not have a fusion. However, as already stated, a fusion was done when it seemed indicated and it would be impossible to state the cost had it not been done.

SUMMARY AND CONCLUSIONS

In a series of 224 patients explored for protruded intervertebral disc, the preoperative clinical diagnosis was 81.25 per cent correct. Of the 224 patients, 103 had preoperative pantopaque studies with an accuracy of 76.7 per cent for that test. We believe pantopaque myelography is a valuable aid in diagnosis and in the past 2 years 71 per cent of the patients whom we explored for protruded intervertebral disc had preoperative pantopaque studies.

The records of the insurance carriers on 88 compensation patients operated upon in this series showed that the workman lost an average of 356.5 working days from the onset of symptoms until he was able to go back to work. Three of the 88 patients were given permanent total disability awards, 8 were closed with no award, and 77 were closed with a permanent partial disability award which averaged $1,015.40. The average cost to the insurance carrier of handling each of the 88 cases was $3,188.75.

A summary of the results upon 160 of the 224 patients, followed from 1 to 10 years after operation, revealed that 61.25 per cent obtained "excellent" results, 28.75 per cent "good" results, and 10 per cent "poor" results.

An excellent or good result is obtained more frequently when a protruded

intervertebral disc is found and removed than when there is a negative exploration or when exploration reveals an inflamed nerve root, adhesions about the nerve root, or some other pathological condition.

Private patients reported better results following operation than did compensation patients.

Spinal fusion need not be done routinely to obtain good results but should be done when indicated. In our series of 224 patients, 37.9 per cent had spinal fusions at the time the protrusion was removed.

CASE REPORTS

GIANT-CELL TUMOR OF BONES OF THE SKULL

REPORT OF TWO CASES

JOHN C. MCNERNEY, M.D.*

Department of Surgery (Neurosurgery), Emory University School of Medicine, Emory University, Georgia

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Benign giant-cell tumors of bone occur most frequently in the long bones and very rarely involve the bones of the skull. While most giant-cell tumors of the skull occur in the mandible (18 cases),[5] 4 cases have been reported in the sphenoid bone,[1,5,11] 2 in the frontal bone,[3,8] 2 in the occipital bone,[5,10] and 1 in the mastoid area which involved the external auditory meatus.[5] The purpose of this paper is to report 2 additional cases of giant-cell tumor of the skull, 1 involving the sphenoid bone, the other the temporal bone.

CASE REPORTS

Case 1. Headaches and failing vision. Amenorrhea. Sella turcica destruction. Craniotomy and partial extirpation. Amelioration of symptoms. Roentgen therapy.

May 30, 1947. Admission of Miss O. E. D., aged 30, a right-handed textile worker with complaints of headaches and failing vision.

History. The patient had had sinus trouble for 2 years. Transient visual hallucinations in the form of bright lights and transient diplopia began 18 months before admission, followed by amenorrhea 6 months later. Failing vision and intermittent olfactory hallucinations of a foul odor had been present for 10 months along with severe frontal and bitemporal headaches. A tonsillectomy and a nasal operation had been performed in October 1946. Since that time her voice had become nasal and she had had an occasional thin pink watery discharge from her left nostril. Although her headaches had decreased in severity, her vision had become progressively worse and she had lost her sense of smell.

Examination. The patient was pale, poorly nourished, and somewhat emotionally labile. Smell was lost on the right and was nearly gone on the left. A thin pink watery discharge escaped intermittently from the left nostril. The right pupil was larger than the left. She had a secondary optic atrophy which had advanced to blindness on the right, and a choked disc with temporal pallor, temporal field cut and 20/40 acuity on the left. There was hypesthesia to pin prick over the left maxilla. The voice had a distinctly nasal quality.

Hematologic examination: r.b.c. 3,580,000, hb. 70.2 per cent, and w.b.c. 7,200. Total protein content of ventricular fluid was 10 mg. per cent. The blood Kahn reaction was negative.

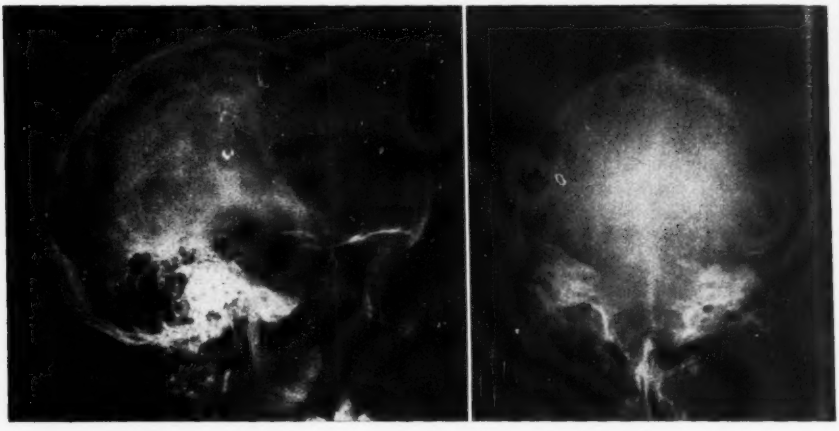
Roentgenograms of skull (Figs. 1 and 2) revealed complete destruction of the dorsum sellae, the floor, and the posterior clinoid processes. The anterior clinoid processes were sharp and pointed, the right smaller than the left. The space between the apices of the petrous pyramids appeared widened. There was a soft tissue mass outlined in the posterior nasopharynx, although this mass was not confirmed on pharyngoscopic examination.

Presumptive diagnosis: Atypical pituitary adenoma.

Operation, May 31, 1947. Under local novocaine anesthesia, supplemented by pentothal sodium, a right frontal 4-burr-hole osteoplastic flap was turned down. The scalp was hinged anteriorly and the flap laterally on the temporal muscle. The dura, which was not particularly taut, was opened and reflected posteriorly. The convolutions were not broadened. A brain

* National Naval Medical Center, Bethesda, Maryland.

cannula was introduced into the anterior horn of the right ventricle. The ventricular fluid was not under increased pressure. Following the right olfactory nerve for orientation, the right frontal lobe was elevated. A bulging mass protruded out of the mesial portion of the posterior part of the anterior fossa. The right optic nerve was flattened, the left not visualized. The mass appeared to be entirely extradural. Dissection was continued back to the anterior rim of the optic chiasm. The diaphragm of the sella turcica was coagulated and incised. Several large biopsy curettements were taken. The tissue had a muddy chocolate color and bleeding became severe, and one wondered if this were not a tumor of some other than pituitary origin. With suction and curette fully 1 oz. of tissue was evacuated. The frozen biopsy section was



FIGS. 1 and 2. Case 1. Roentgenograms demonstrating destruction of dorsum sellae, floor and posterior clinoid processes.

reported as being a giant-cell sarcoma. The cavity was packed with oxycel strips. Bleeding had been voluminous. Although the patient had received 2 whole blood transfusions during the procedure, her condition remained extremely critical. Consequently she was given a 3rd transfusion of whole blood intra-arterially, following which her blood pressure rose to a normal level and she was returned to her room in satisfactory condition.

Pathologic Histology. The tissue consisted of a fibroblastic matrix which contained large multinucleated giant cells with prominent nuclei. A diagnosis of benign giant-cell tumor of bone was made (Fig. 3).

Subsequent Course. The patient made an excellent operative recovery. She received 300,000 units of penicillin each 24 hours for the first 10 postoperative days. During her convalescent period she remained free from headaches and left rhinorrhea. On discharge June 17, 1947, following roentgen therapy, 1200 R to each 8 cm. bitemporal portal at 50 cm. target distance, the left temporal field defect was no longer present and she could read newspaper print.

2nd Admission, Sept. 24, 1947. She re-entered the hospital for a 2nd course of x-ray therapy. Her general physical and mental condition were excellent. Amenorrhea persisted and she had had an intermittent left rhinorrhea. Examination revealed a well healed right frontotemporal craniotomy scar. Visual acuity was not tested and refractive corrections were not made. Water metabolism was normal. Glucose tolerance test was normal. The basic metabolic rate was −22 per cent. The condition of her skin did not warrant further roentgen therapy. A plastic repair of her spinal fluid fistula was likewise deferred at this time and she was discharged on September 27 after being placed on sulfadiazine and thyroid.

Case 2. Head trauma. Throbbing left temporal pain. Left temporal mass. Destruction of left zygoma. Aspiration biopsy. Left external carotid ligation. Left temporal craniotomy and extirpation. Amelioration of symptoms. Roentgen therapy.

Aug. 28, 1946. Admission of Mr. C. H. McM., aged 39, a saw-mill worker with complaints of throbbing left temporal pain, left tinnitus, and impaired left hearing.

History. The patient had had chronic sinus disease which had required surgical drainage on the left some years before. Four years before admission he had been rendered unconscious in an automobile accident when he received a severe blow across the bridge of his nose and under his chin. For 1 year he had had throbbing left temporal pain associated with a pulsating roaring in the left ear. Hearing in the left ear had become progressively impaired and he had had occasional dizziness. For 4 months he had noticed a left temporal mass.

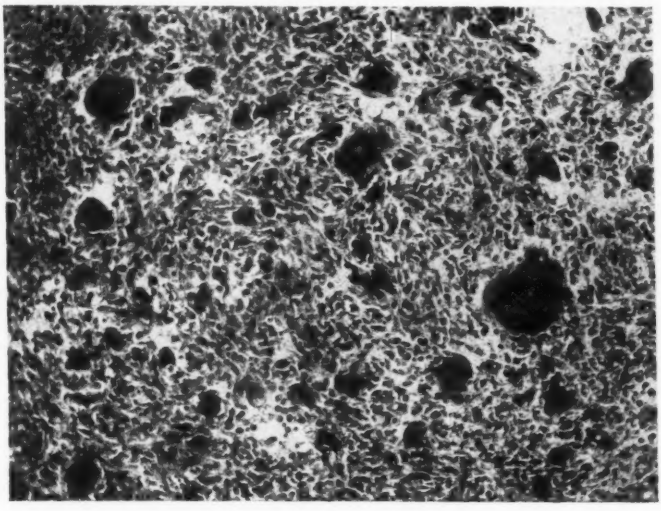


FIG. 3. Case 1. Photomicrograph of tissue removed at operation showing numerous large multinucleated giant cells. Hematoxylin and eosin (×150).

Examination. The patient was well developed and well nourished. A rather firm, tender, slightly fluctuating mass was present in the left temporal area anterior to the ear. The posterior portion of the left zygoma was enlarged. The anterior superior wall of the left external auditory canal bulged. Hearing was markedly reduced on the left, with loss of air conduction.

Hematologic examination: r.b.c. 4,640,000, hb. 88.4 per cent, w.b.c. 12,550. The sedimentation rate was normal; the blood Kahn reaction negative.

Roentgenograms revealed a slight clouding of the left mastoid structure without bony change, no evidence of paranasal sinus disease, and an area of destruction of the left temporal bone and the posterior portion of the left zygoma.

On *Sept. 7, 1946* an aspiration biopsy was done and 8 cc. of brown fluid which contained small portions of tissue were withdrawn. Histologic examination was not conclusive. Because of the extent of involvement of the left zygoma and the presumptive malign character of the lesion, the patient was given roentgen therapy: 800 R to a 6 cm. left temporal portal at 50 cm. target distance. On *Sept. 13, 1946* a 2nd biopsy was performed and the patient was temporarily discharged.

Pathologic Diagnosis. Benign giant-cell tumor.

2nd Admission, Oct. 30, 1946. The patient re-entered the hospital for operation. In the interval since his discharge, left temporal headache had been almost constantly present. Except for an increase in the size of the posterior portion of the left zygomatic arch, the objective findings remained unchanged. The plasma alkaline phosphatase level was 4.1 Bodansky units; serum calcium 10.5 mg. per cent; and serum phosphorus 2.9 mg. per cent.

Roentgenograms of the skull (Fig. 4) revealed an increase in destruction of the inner and outer tables of the squama of the left temporal bone, which extended anteriorly to the posterior margin of the greater wing of the left sphenoid bone and posteriorly to the anterior margin of the left parietal bone.

FIG. 4. Case 2. Lateral roentgenogram of skull showing large defect of left temporal bone.

On *Oct. 30, 1946*, under local novocaine anesthesia, the left external carotid artery was ligated.

Operation, Oct. 31, 1946. Under local novocaine anesthesia supplemented by pentothal sodium, a horse-shoe incision was made from the anterior margin of the left zygoma over the temporal bone to just behind the left ear. The scalp was reflected, temporal muscle incised, and the fascia subperiosteally dissected upward. In the posterior inferior portion of the temporal bone a yellow domed tumor mass had broken through. It had not invaded the muscle. Four burr openings were made and were connected with the De Vilbiss in such manner that the mass was circumscribed by healthy bone except at its most inferior level, which was the zygoma. The tumor was incised and cored. It contained yellow fluid that might well have had cholesterol crystals in it. The growth was rather firmly attached to the dura, which it did not invade. After coring, the wall was taken down to the floor of the middle and anterior fossae. There were two breaks in these bones, one into the left frontal sinus and orbital cavity, and a posterior one which emptied into the left auditory canal. A biopsy was made. The pathologist reported that the tissue was not cholesteatoma but a giant-cell tumor. At this stage the parasellar portion of the sphenoid bone had been reached and rather than get into too dangerous territory, especially since it appeared doubtful that a permanent cure

could be effected, the openings in the floor of the fossa were sealed with gelfoam strips after the tumor bed had been zenkerized. Four gm. of sulphanilamide crystals were introduced into the bottom of the wound. A gelfoam pack was used to obliterate dead space. The temporal muscle was sutured in 3 layers and the scalp in 2. The patient was given a transfusion of 500 cc. of blood and left the operating room in satisfactory condition.

Pathologic Histology. Sections revealed a dense fibroblastic matrix within which were numerous areas of multinucleated giant cells. Diagnosis: Benign giant-cell tumor of bone (Fig. 5).

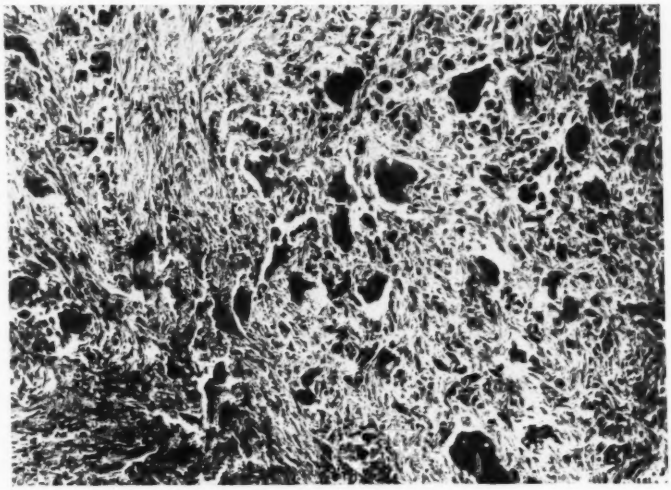


FIG. 5. Case 2. Photomicrograph of the fibroblastic tissue removed at operation. Note numerous multinucleated giant cells. Hematoxylin and eosin (×125).

Subsequent Course. The patient made an excellent operative recovery. A left rhinorrhea ceased permanently 48 hours after operation. On discharge Nov. 13, 1946 his left temporal headaches were improved, and left tinnitus had disappeared. A peripheral left facial paresis and a left otorrhea, for which he received sulfadiazine, persisted.

Six weeks later roentgen therapy was given: 1200 R to an 8 cm. left temporal portal at a 50 cm. target distance. Six months after operation the patient was symptom free and his left otorrhea had ceased. A mild residual left facial paresis persisted. At present he has no complaints, feels well, and has been back at work for 6 months. Roentgenograms on Sept. 30, 1947 revealed that the contour of the bony defect in the squamous portion of the left temporal bone was smoother and more distinct.

COMMENT

The infrequency of occurrence of giant-cell tumors in bones of the skull needs no further comment. These 2 reports increase to 11 the recorded cases that involve bones of the skull other than the mandible.

Points of interest in Case 1 include the absence of a history of trauma and the fact that with complete destruction of the sella turcica, primary tumor of bone was not considered as a presumptive diagnosis although the malign behaviour at times of benign giant-cell tumor to destroy bone is well recognized.[2,4,6,7,9]

In Case 2, the location of the lesion is most unusual. The medical literature failed to reveal another case of giant-cell tumor of the temporal bone. The cystic character of the lesion and the fact that many giant-cell tumors of bone are manifestations of hyperparathyroidism stress the need for blood chemical studies in similar cases.

Giant-cell tumors arise from resorptive processes in cartilaginous bone with the possible exception of the type that Geschickter and Copeland[5] refer to as subperiosteal. This theory fails to account for the occurrence of such a tumor in the squamous portion of the temporal bone (membranous bone), a fact that has been commented upon by Keegan and Baker[8] in their report of a giant-cell tumor of the frontal bone.

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NON-TRAUMATIC ATLANTO-AXIAL DISLOCATION

REPORT OF CASE WITH RECOVERY AFTER QUADRIPLEGIA

Leonard A. Titrud, M.D.,* C. A. McKinlay, M.D., Walter E. Camp, M.D., and Hewitt B. Hannah, M.D.

Minneapolis, Minnesota

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In 1830, Bell[1] described the occurrence of dislocation of the atlas because of destruction of the transverse ligament that holds the odontoid process of the axis in its normal anterior position. This particular instance was associated with a neck infection in the patient. Greig[8] demonstrated that during a cervical infection, a diffuse tissue hyperemia occurs which results in adjacent bone decalcification with loosening of the intervertebral ligaments, especially between the atlas and axis, so that the skull and 1st cervical vertebra dislocate anteriorly. In 1932, after a thorough study of this disease process, Jones[15] emphasized specifically that dislocation results because the decalcified arch of the atlas no longer will permit a firm attachment of the

* 1141 Medical Arts Bldg., Minneapolis 2, Minnesota.

transverse ligament which is chiefly responsible for holding the odontoid process anteriorly against the atlas.

At this time, Jones[15] reviewed 14 cases in the literature and added 2 personal instances. By 1934, Jones and Roberts[16] presented 5 additional cases bringing the total to 21. It was learned that this disease occurred most frequently between the ages of 6 and 12 years and was distributed equally between the sexes.[15] Almost all of the individuals had a precedent inflammatory condition of the neck such as: abscess, tuberculous cervical glands, or syphilitic pharyngeal ulceration. Fitzwilliams[7] had an instance occurring with acute rheumatism; Böger,[3] Ely,[4] Stammers and Frazer,[13] and Evans[6] reported additional cases associated with rheumatic fever. Trauma is not a principal factor. The type of infecting organism made no difference since the hyperemia throughout the cervical tissues was directly responsible for the decalcifying process. However, Berkheiser and Seidler[2] believed that the streptococcus was the most common organism in their 5 cases, which are included with the series of Jones and Roberts.[16] Approximately 10 days after the throat infection, a sudden neck pain or torticollis was usually experienced. At this time, cervical x-rays revealed a forward dislocation of the atlas on the axis with vertebral decalcification, especially of the arch of the atlas.[15] The usual treatment has been manipulative reduction and traction fixation or plaster immobilization for approximately 10 weeks.[15] By this time, recalcification has occurred.

Jones and Roberts[16] provided a logical clinical classification of this syndrome:

First Type. This is the most common. The atlas is dislocated forward and rotated slightly but enough to produce torticollis with painful movement and deep cervical muscle spasm.

Second Type. The displacement is more marked, with spinal cord compression but with survival of the patient. Jones and Roberts[16] reported the first case of this type, which was that of a quadriplegic child who made a full recovery after reduction of the dislocation. Our adult patient, who had complete recovery of quadriplegia following surgical spinal cord decompression, fits into this group.

Third Type. The dislocation is complete, with sudden death from spinal cord compression, such as in the cases of Bell,[1] Greig,[8] Reid,[12] and Jones.[16]

In 1935, after a thorough search of the literature, Hess, Bronstein, and Abelson[10] found a total of 22 cases and added 2 of their own. Although this is mainly a disease of children, they found mention of it in only one pediatric journal.[14] They emphasized the avoidance of excessive rotation and manipulation of a child's head during surgical drainage of cervical infections since the relaxed anesthetic muscles may not protect against vertebral dislocation. These same authors[9] reported in 1942 a 5-year follow-up of a 12-year-old boy who had an unreduced spontaneous atlanto-axial dislocation. Their suggestion was that although occasionally spontaneous reduction and normal calcification may occur, some of the persisting spinal deformities in older persons may have arisen as in their described case. Martin[11] added 4 personal cases to the literature in 1942. These instances were in children without neurological complications. In the same year, Englander[5] presented a girl with this syndrome accompanied by neurological damage from an injured spinal cord.

Our case reported here is of an adult who developed quadriplegia from spinal cord compression and recovered after surgical decompression.

CASE REPORT

Mrs. R. R., a 40-year-old white housewife, was first admitted to the Northwestern Hospital on Sept. 20, 1946. She had had a sore throat and stiff neck since Aug. 1, 1946. Polio-

myelitis had been suspected, but the spinal fluid had proved to be normal. Two weeks' treatment with penicillin and a sulphonamide was given at another hospital during August and September. On Sept. 20 she experienced a full sensation in the throat besides occipital pain, with difficulty in raising sputum and swallowing solid food.

Examination. Blood pressure 162/108; pulse 70–110; respirations 20; oral temperature 99.2°. Hb. 77 per cent; rbc 4,442,000; wbc 12,100: 89 per cent polymorphonuclears; 11 per cent lymphocytes. Sedimentation rate, 96 mm. in 1 hour. The essential physical findings were a diffusely edematous appearing pharynx, stiff neck, and non-tender cervical lymphadenopathy.

Course. It was believed that a retropharyngeal abscess was present, and this was incised and drained Sept. 20. Unfortunately, the pus was not cultured. From Sept. 20 to 26, 40,000 units penicillin were given intramuscularly every 3 hours, in addition to warm saline throat

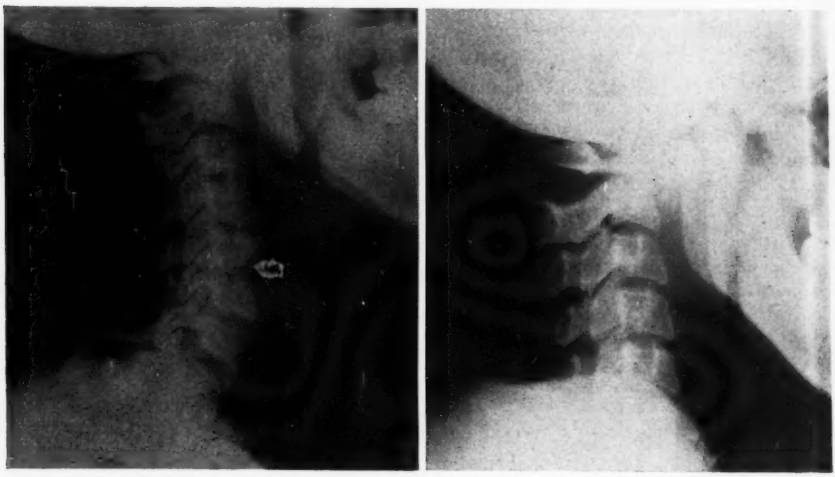

FIG. 1 (*left*). Oct. 1, 1946. Lateral cervical spine revealing postpharyngeal swelling and anterior displacement of 1st cervical vertebra on the 2nd.

FIG. 2 (*right*). Oct. 15, 1946. Lateral cervical spine showing increase of retropharyngeal swelling with vertebral displacement and constriction of spinal canal. Odontoid process destruction exists.

irrigations. Temperature ranged from 99.8° on Sept. 21 to 98.6° on Sept. 26. The wbc on Sept. 23 was 7700 per c. mm. She persisted in complaining of a stiff and painful neck, and was discharged on Sept. 27, 1946.

2nd Admission. Because of continued pharyngeal infection, she was hospitalized Oct. 10, 1946 for streptomycin therapy. Oral temperature was 98.6°. On Oct. 11, 10 cc. of pus were evacuated from the pharyngeal abscess on the left side of the throat. No culture was made. From Oct. 11 to 13, 20,000 units penicillin were given intramuscularly every 3 hours. From Oct. 13 to 15, 100 mg. streptomycin were given intramuscularly every 3 hours. On Oct. 14, the neck pain was severe and numbness of the left fingers and weakness of the left arm developed. By Oct. 15, the right fingers had become numb and the right arm was weak. Oral temperature was 99.5°.

8:00 A.M. Oct. 15. Examination revealed absent vibration sense in the fingers of both hands. There was marked weakness of the left upper extremity and less paresis of the left lower. Abdominal reflexes were absent and there was a left Babinski sign. Deep reflexes were

equal except for a reduction of those in the left arm. Bilateral ankle clonus was present.

4:00 P.M. Oct. 15. Examination again revealed the cranial nerves to be normal. There was almost complete quadriplegia except for slight movement of right arm and leg. The entire body was drenched with cold perspiration. Deep reflexes in the upper extremities seemed fairly active and equal. Patellar and Achilles reflexes were equally hyperactive. There were bilateral ankle clonus and equivalent Babinski signs. Cutaneous hyperesthesia existed everywhere below the head. The bladder had a urinary retention of over 1000 cc.

The cervical spine x-rays of Oct. 15 were compared with those of Oct. 1, and the postpharyngeal soft tissue swelling was seen again (Figs. 1 and 2). The x-rays of Oct. 15, however, revealed an anterior displacement of the 1st cervical vertebra on the 2nd, with destruction of the odontoid process. It was feared that the spinal cord compression was caused by an osteomyelitic vertebral displacement together with an epidural abscess. Therefore, it seemed that an immediate cervical cord decompression should be done.

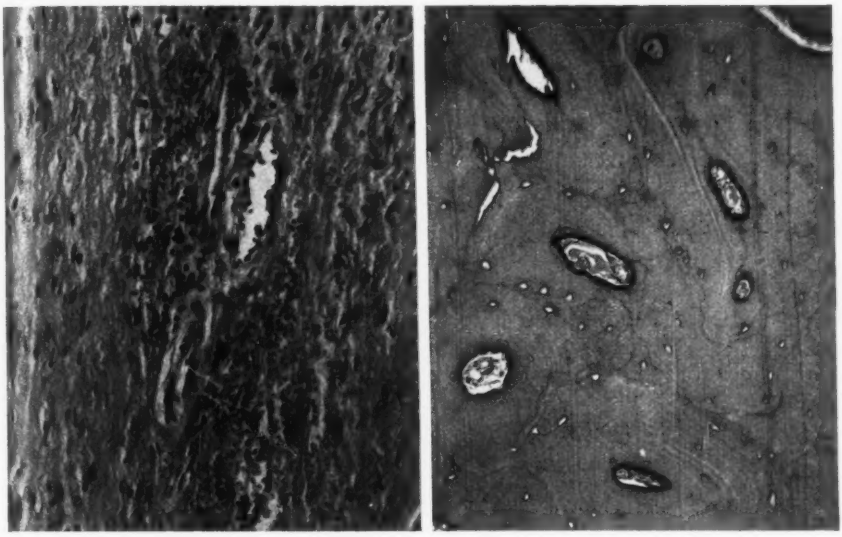

FIG. 3 (*left*). Section of extradural tissue removed Oct. 15, 1946. Inflammatory reaction with perivascular lymphocytic infiltration throughout fibrous tissue exists.

FIG. 4 (*right*). Section of decalcified bone removed from atlas Oct. 15, 1946. This bone was soft. No inflammatory reaction seen.

6:30 P.M. Oct. 15. Operation. Anesthetic: intratracheal nitrous oxide and oxygen with intravenous pentothal and curare. During the procedure 500 cc. saline and 1000 cc. blood were given intravenously. With the patient in a prone position and the face supported by a cerebellar head rest, a bilateral laminectomy of the 1st, 2nd, 3rd, and 4th cervical vertebrae was done together with a generous decompression of the foramen magnum by removal of occipital bone. There seemed to be at least 1 cm. of anterior displacement of the atlas on the axis. There was a marked angulation and compression of the cervical dural sac from the 1st to 3rd vertebral level. In this area, there was an extradural layer of edematous and firm tissue 3–4 mm. thick; after removal of this, the dural sac began to have visible pulsations. [Subsequent microscopic study (Fig. 3) of this extradural tissue revealed moderate inflam-

matory reaction.] The bone of the 1st and 2nd cervical vertebrae seemed soft and necrotic (Fig. 4). No actual pus was encountered. Cultures of the wound were sterile. The dura was not opened. After the spinal cord was decompressed and its angulation obliterated, the wound was closed and sterile dressings were applied. A posterior molded plaster cast was fitted over the head, neck, and all of the back. The procedure occupied 3¾ hrs. and was tolerated well.

Course. Her temperature decreased gradually from 100.8° on Oct. 16 to normal on Oct. 20. From Oct. 16 to 27 she was given 25,000 units penicillin intramuscularly every 3 hours. On Oct. 16, right arm and leg movement began. By Oct. 19, left arm movement began, and on Oct. 20, superficial cutaneous sensibility seemed normal everywhere and little impairment of deep sensibility persisted. At this time, biceps and triceps reflexes had become quite active and equal, and patellar and Achilles reflexes were equally hyperactive bilaterally. There was unsustained left ankle clonus. Babinski tests caused severe pain. Her retention catheter was removed Oct. 21, after which no urinary disturbance existed. By Oct. 25, she was still unable to hold objects in her hands although strength was returning gradually.

On Oct. 30, an anterior molded plaster cast was applied from the neck to the pubis, and on Nov. 2 it was incorporated into a complete Minerva cast. By Nov. 5, she sat up for the first time. On this date, reflexes in all extremities were hyperactive, with bilateral ankle clonus. Paresis in the left arm still persisted. Roentgenograms of cervical spine revealed no change in the bony destruction. She was discharged to her home Nov. 8, 1946.

3rd Admission, Feb. 1–3, 1947. A window was cut in the posterior cervical area of the cast. The wound was well healed. Cervical spine x-rays Feb. 3 revealed a slight increase in the anterior displacement of the body of the 1st on the 2nd cervical vertebra. Urinalysis and blood count were normal. Her principal complaint was of some numbness in the finger tips of both hands. There was good muscular power in all extremities. The patient had been working at home and getting along quite well. Because of the fear that cervical infection had existed, spinal fusion was postponed.

4th Admission, Mar. 20, 1947. The cast which she had worn since Nov. 2, 1946 was bivalved.

Operation, Mar. 21, 1947. Spinal fusion. Anesthetic: intratracheal nitrous oxide and oxygen and intravenous pentothal and curare. During the procedure 500 cc. saline and 750 cc. blood were given intravenously. The patient was in a prone position with her face supported on a cerebellar head rest. An exposure was made of the occipital edges of the enlarged foramen magnum. The scarred tissue over the posterior surface of the dura as well as the adjacent bony wall of the vertebral canal was exposed from the occiput to the 5th cervical vertebra. The laminae of the 5th and 6th cervical vertebrae were also denuded of soft tissue. Cultures from the wound were sterile. Bone grafts were taken from the outer table of the left posterior iliac wing. Two grafts were secured, each measuring 16 cm. long, 2 cm. wide, and 5 mm. thick. These were well curved and conformed to the occipital surface, with the long shank of each graft lying along the posterior cervical canal. Bone chips were placed about the grafts. The sutured overlying soft tissues served to fix the grafts in place. A posterior molded plaster cast was finally applied over the head, neck, and entire back. The procedure took 3 hrs. and was tolerated well.

Course. Convalescence was satisfactory and the wound healed well. From Mar. 21 to Apr. 1, 50,000 units penicillin were given intramuscularly every 3 hours. The patient was immobilized in a plaster shell until a cervical brace was fitted for her in April. X-rays of cervical spine Apr. 21 (Fig. 5) revealed no change in vertebral alignment from that of Feb. 3. The upper part of the graft under the occiput was fractured. She was discharged to her home Apr. 26 and informed to wear the brace both day and night continuously.

She returned to work about her home. Her chief distress concerned pain behind each ear which was believed to have been caused by injury of the cervical nerves. Her weight increased from 101 pounds in April to 120 on July 28, 1947. At this time, neurological examination was negative. Cervical x-rays showed no change in relationship between the atlas and axis. There was increased bone deposition about the graft.

The last examination was Nov. 6, 1947. She weighed 130 pounds and was worried about

overweight. The postauricular pain was gone. A slight numb and tight sensation persisted in the finger tips of both hands. She was able to perform intricate movements with her fingers and practised her hobby of painting pictures satisfactorily. There was only a slight increase in the left biceps and triceps reflexes. X-rays (Fig. 6) showed no essential change from those of July. The wearing of the cervical brace was discontinued at this time, after 6½ months' use and 7½ months after the spinal fusion. She had made a quite complete recovery, approximately 13 months after the incidents of the pharyngeal abscess, hyperemic decalcification and dislocation of the atlas on the axis, and quadriplegia.

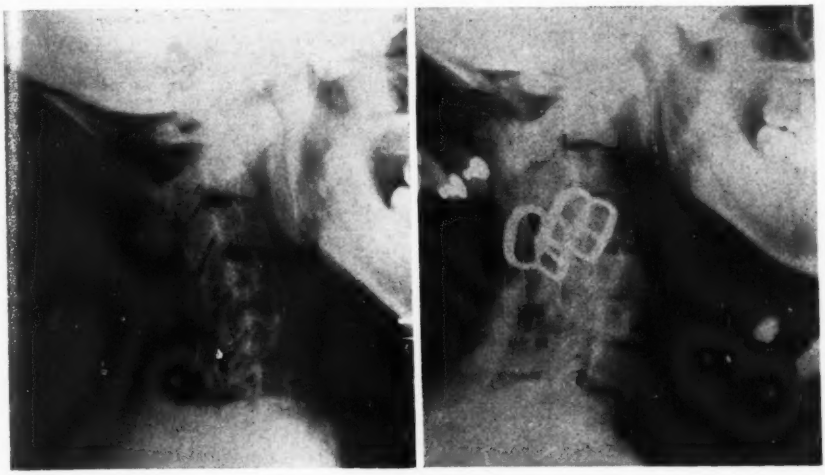

FIG. 5 (*left*). April 21, 1947. Lateral cervical spine after decompression of foramen magnum and cervical spinal cord by laminectomy of upper 4 cervical vertebrae. Retropharyngeal swelling now absent.

FIG. 6 (*right*). Nov. 6, 1947. Lateral cervical spine revealing spinal fusion. Patient was asymptomatic and ready to discontinue use of cervical brace.

SUMMARY AND CONCLUSIONS

1. Attention is called to a rare syndrome wherewith infection about the mouth or neck provokes deep cervical hyperemia with resultant adjacent vertebral decalcification, loosening of attached ligaments, and spinal dislocation.

2. An instance occurring in an adult is described who developed quadriplegia from vertebral dislocation and spinal cord compression, and recovered completely after surgical decompression.

A warning is issued to be aware of this condition, especially in children who have neck pain or torticollis in the presence of throat infections.

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EXTRADURAL HAEMORRHAGE AS A CAUSE OF CORTICAL BLINDNESS

A. N. Guthkelch*
Manchester, England

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A survey of some recent relatively large series of cases of extradural haemorrhage[1,2,3,4] suggests that about 5 to 10 per cent of them all are found in the occipital region. But of the references cited McKenzie[2] remarks specifically that visual field changes, in his experience, did not occur in extradural haemorrhage, while neither Gurdjian and Webster[1] nor Rowbotham[4] make any mention of such a symptom. Pringle,[3] on the other hand, reported one case of left homonymous hemianopia† occurring in a patient with a right occipital extradural haemorrhage, and the present case, which is one of practically complete cortical blindness due to a bilateral clot, demonstrates a further possible clinical picture, and one that appears not to have been recorded before.

CASE REPORT

Miss L. K., aged 67 years, fell in the street striking the back of her head on April 23, 1947. She was admitted to Hospital next day in a drowsy and irritable condition, and roentgenograms showed a fissured fracture extending upwards from the left margin of the foramen magnum to the lambda. It crossed the left lateral sinus about 2 cm. from the midline. After 4 days her conscious level began to improve, but she complained of blindness, though it was thought that as the days passed she was beginning to distinguish light from darkness.

* 8 St. James' Square, Manchester 2, England.

† Whether or not macular vision was spared is not recorded.

The patient was first seen by the present writer on May 8th. She was then garrulous and euphoric and quite devoid of insight into her condition. She knew the date and day of the week, but was able to say where she was only after some hesitation. She was reported to be incontinent of urine. On testing her visual acuity it was found that she could dimly perceive hand movements in all parts of the visual field of both eyes, but could not count fingers. The optic discs both showed acute papilloedema with haemorrhages but no exudates. The pupils

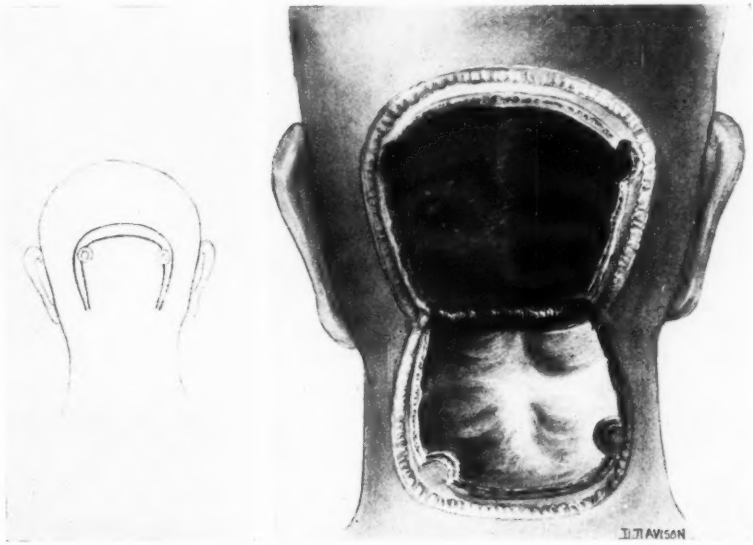

FIG. 1. A semidiagrammatic sketch showing appearance of the dura mater after removal of the clot. (Redrawn by Miss D. Davison from sketch made by the writer at operation.)

were equal and of normal size, and they reacted briskly to light and on convergence. The remainder of the neurological examination was negative.

The physical signs were obviously those of an almost complete bilateral interruption of the visual pathways at a "supranuclear" level, and it was thought that the most likely diagnosis was one of bilateral occipital subdural haematoma.

Operation. On May 10, 1947 bilateral occipital burr holes were made, as though for ventriculography. These both revealed a subjacent mass of black semi-solid extradural clot. The incisions were joined and extended downwards in the form of a small osteoplastic flap hinged on the suboccipital muscles and exposing the upper 2 cm. of the dura of the posterior fossa (Fig. 1). It was now possible to remove from the extradural space a huge mass—some 120 cc. in all—of black clot mixed with brownish syrupy fluid, leaving the dura covered with rather tough brown granulation tissue. We were unable to identify with certainty the exact site of the original haemorrhage, but it presumably occurred from somewhere in the region of the torcular.

The dura was now hitched to the pericranium with a number of interrupted sutures, the bone flap replaced and the wound closed in the usual manner. At the end of the operation the patient was able to count fingers easily.

Progress. During the next 2 days she became mentally much clearer, and ceased to be

incontinent. Her visual acuity improved so rapidly that she was able to read a newspaper comfortably within 4 days of operation. She was discharged from Hospital on the 11th day. The papilloedema was then rapidly subsiding and there was no remaining diminution of visual acuity, nor were there any field changes.

When last seen, in March, 1948, the patient was in excellent health, and reported that she was able to thread her needle and do other fine work just as well as before her accident. Her visual fields remained full and her optic discs had returned to normal.

COMMENT

It would indeed be a strange thing if the visual cortex were for some reason immune from the effects of compression by an extradural haemorrhage, and there is no reason to suppose that McKenzie[1] for example, intended to draw this inference. The explanation of the lack of records of visual field changes in extradural haemorrhage is presumably that in most cases the patient is too acutely ill, and at too low a conscious level to permit of accurate testing. However, from time to time as in the present instance, cases are seen in which the clot either develops more slowly or ceases to grow before causing fatal compression, so that although there is evidence of focal brain damage, the conscious level remains or returns to normal. It would therefore be interesting to complete the picture by finding an example of a hemianopia occurring in an extradural haemorrhage of the temporal region, just as it may occur in tumours of the temporal lobe of the brain.

SUMMARY

A case of almost total cortical blindness due to a bilateral occipital extradural haematoma is recorded. Vision was completely restored by operation.

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MENINGIOMA OF THE SPHENOID RIDGE IN A CHILD

Collin S. MacCarty, M.D., and Louis J. Gogela, M.D.*
Section on Neurosurgery, Mayo Clinic and Mayo Foundation, Rochester, Minnesota

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The fact that meningiomas tend to occur most commonly between the ages of 35 and 55 serves as a deterrent to their inclusion in the preoperative differential diagnosis of intracranial lesions appearing during childhood. Meningiomas in children have been reported frequently and have usually been found lying over the

* Fellow in Neurosurgery, Mayo Foundation, Rochester, Minnesota.

cerebral convexities or within the longitudinal cerebral fissure. Frazier and Alpers[3] reported 6 patients ranging in age from 3 to 18 years with such lesions in a series of 75 meningiomas. The youngest patient in the group of cases published recently by Grant[4] was 2 years old.

However, meningiomas arising along the sphenoid ridge have not been encountered in patients as young as those just cited. Cushing and Eisenhardt[1] in their series of cases of meningiomas along the sphenoid ridge, reported their youngest patient to be 26 years old. In the series of such lesions published by Groff,[5] the youngest patient was a Negress also 26 years of age. Elsberg, Hare and Dyke[2] described an instance of exophthalmos due to a meningioma, probably arising from the sphenoid ridge, in a 19-year-old girl. The case that we are reporting is that of a boy 12 years of age who had had ocular proptosis since the age of 5. Mucocele was considered the most likely cause of the ocular protrusion; hence, the first surgical procedure was planned with this diagnosis in mind.

REPORT OF CASE

A boy 12 years of age, registered at the Mayo Clinic on April 10, 1947. His chief complaint was proptosis of the left eye associated with pain about the eye.

History. Data concerning his birth, development and family were all irrelevant. He was in the sixth grade, was a good student and enjoyed sports. His parents first noted protrusion of his left eye when he was 5 years old. The degree of protrusion appeared to vary from time to time, but it was gradually progressive. Swelling of the lid of the left eye occurred with colds. During succeeding years proptosis was asymptomatic and the boy led a full, normal life. At the age of 8 years, the lad struck his head on falling off a pony. Headache developed subsequently and the patient was comatose for an hour. No apparent residua or sequelae followed this accident.

About 3 months prior to registration, headache became a prominent symptom. The patient described the ache as being of the "pressure" type and stated that it usually occurred in the early morning. At times, he was awakened by pain. It was most intense about the left eye and extended posteriorly into the occipital region. At the time of initial onset, the headaches were relatively mild, occurring about once a week and lasting a few hours. The pain progressed in severity and frequency and became a more or less steady ache which was aggravated by coughing and accompanied by nausea and vomiting during the 10 days before examination. It became sufficiently intense to warrant use of narcotics to obtain relief.

Examination. The boy was well developed and well nourished. Except for obvious proptosis, results of general physical examination were entirely negative. Blood pressure measured 120 mm. of mercury systolic and 50 diastolic; pulse was 70. There were no abnormal neurologic findings. The right eye appeared normal. The left eye protruded and was displaced downward and laterally. The exophthalmometer measured proptosis to the extent of 4 mm. The external ocular muscles were not noticeably embarrassed and tension in the eye was normal. The pupils were bilaterally equal in diameter and were active. The fundi were normal. Vision in the right eye was reported at 6/12, in the left eye at 6/30, without the corrective lenses which the patient was accustomed to wearing. Otorhinolaryngologic examination disclosed only slight mucopurulent nasal discharge. Urine and blood findings were all within normal limits and results of flocculation and tuberculin tests were negative.

Roentgenograms of the head and orbits revealed a large mass in the left fronto-ethmoid region which had eroded and decalcified the supra-orbital ridge, the roof of the orbit, and the greater wing of the sphenoid and had, in addition, visibly expanded intracranially (Fig. 1). Special roentgenographic studies further disclosed diffuse opacity of all paranasal sinuses. Roentgenograms of the thorax were negative.

Operation 1. As said previously, mucocele was considered the most likely lesion and surgery was directed with this diagnosis in mind on April 23, 1947. With the patient under

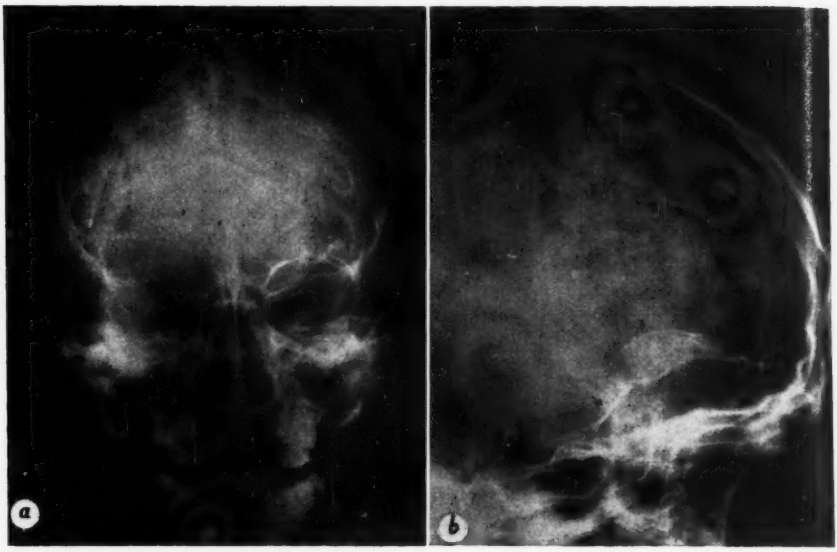

FIG. 1 (*a*) Preoperative anteroposterior roentgenogram, showing a calcified tumor in left fronto-ethmoid and frontosphenoid regions. (*b*) Preoperative lateral roentgenogram of tumor, same date.

FIG. 2. Postoperative roentgenogram following removal of tumor, orbital roof and involved portion of the sphenoid ridge.

general anesthesia, the usual type of external frontal incision was made on the left; more bleeding was encountered than usual. The periosteum was reflected and a defect was encountered in the superior orbital plate in the floor of the left frontal area. The presenting tumor mass appeared more vascular than a mucocele and was immediately assumed to be a meningioma. Microscopic examination of a frozen section of tissue removed for biopsy confirmed the suspicion. Bleeding was controlled with packs which were removed after 48 hours and replaced with a bit of absorbable gauze. The patient received 250 cc. of whole blood during the procedure.

Following operation, 160,000 Oxford units of penicillin were administered daily by the intramuscular route. The postoperative course was uneventful, and the patient was dismissed on the 12th postoperative day.

Operation 2. The patient's condition remained unchanged. Left transfrontal craniotomy (Fig. 2) was performed on June 11.

1947, with the patient under the influence of open drop ether anesthesia. The tumor was first encountered with the drilling of the anterior temporal burr hole. The bone in this area was invaded by tumor tissue. After removal of the bone flap, the meningioma was recognized presenting between the left frontal lobe and the tip of the temporal lobe. It was large, involving the supra-orbital ridge, the roof of the orbit, the sphenoid ridge, the ethmoids and the anterior clinoids. On piecemeal resection, the mass was found to be attached to the

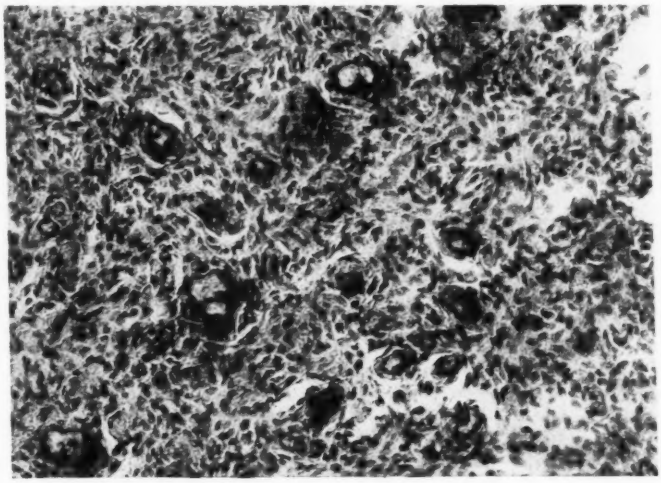

FIG. 3. Section of a psammomatous meningioma. Small, irregular, hyalinized and calcified plaques are numerous (hematoxylin and eosin, ×200).

periosteum of the orbital roof and to the crista galli. Involved portions of the greater wing of the sphenoid, the crista galli, the orbital plate, the posterior wall of the frontal sinus, and a segment of the frontal bone were removed by use of the rongeur. The tumor was removed in its entirety. The periorbita was opened to allow further decompression of the orbit. A gauze pack overlying oxidized cellulose was utilized to control bleeding from the tumor bed. The dura was closed with interrupted sutures. During the course of the operation the patient received 2,000 cc. of whole blood by transfusion.

Course. Postoperatively, 240,000 Oxford units of penicillin were administered daily by the intramuscular route. The pack was removed on the 4th day and the patient was dismissed from the hospital on the 14th postoperative day. According to the exophthalmometer, proptosis of the left eye was 3 mm. at that time.

On re-examination 6 months later, the lad was bright and cheerful and had gained weight. His parents remarked that he was a "new boy around the house." He had been asymptomatic since operation except for 3 or 4 minor headaches each lasting about ½ hour. He had had no convulsions and no motor or sensory disturbances. Eye movements, fundi and visual fields were all normal. Vision was reported to be 6/15 in the left eye and 6/10 in the right with the patient's corrective lenses. There was still a slight persistent downward and forward displacement of the left bulb.

Pathology. On microscopic examination the tumor was recognized as a meningioma of the psammomatous variety. Whorling was absent, but numerous, small, irregular, hyalinized and calcified plaques were present (Fig. 3).

SUMMARY

Although meningiomas of the cerebral convexities are not uncommon in children, their origin from the sphenoid ridge is unusual in a child. Ocular proptosis in the younger age group would lead one to suspect presence of a lesion other than a meningioma.

A case of unilateral exophthalmos in a 12-year-old boy has been presented. Erroneously, initial surgical attack was directed toward the removal of a mucocele. On subsequent intracranial approach a psammomatous meningioma was identified along the sphenoid ridge. Following resection of the tumor mass and the involved bone, the patient made a complete and uneventful recovery.

The case presented may be that of the youngest patient on record with such a tumor. Meningioma of the sphenoid ridge should always be considered a possible cause of proptosis in a child.

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INTRATHECAL ALCOHOL IN THE TREATMENT OF SPASTIC PARAPLEGIA

IRVING S. COOPER, M.D.,* AND THOMAS I. HOEN, M.D.†
Neurosurgical Section, U. S. Naval Hospital, St. Albans, L. I., N. Y.

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THE use of alcohol to produce more prolonged blocking of nervous impulses than infiltration with procaine can afford is a well established therapeutic measure. It has received practically universal use in the infiltration of the gasserian ganglion[7] in patients with trigeminal neuralgia. Paravertebral injection of alcohol as an agent to produce prolonged sympathetic nerve blocks is also a widely used procedure,[2,11] as is the infiltration of peripheral nerves in persistent neuralgias,[3] particularly those of pure sensory distribution. Adson[1] and others have reported the use of alcohol instilled in minute quantities into the subarachnoid space to block particular dorsal roots in the treatment of persistent pain. Until very recently, however, the injection of alcohol in relatively large quantities into the subarachnoid space, for the purpose of relieving the distressing spasticity in the lower extremities in cases of spastic paraplegia, was a relatively unknown procedure. Shelden and Bors[10] recently reported their experience with this measure, and stated that "the amounts of alcohol injected into the subarachnoid space have never been sufficient to control spasticity, except for the recent work of Pudenz and Nourse, whose data are soon to be published." Shelden and Bors reported the use of intrathecal injection of absolute alcohol with immediate relief of spasticity and mass reflexes in a series of 24 patients with spinal cord injuries. The present authors wish to report their results with the use of subarachnoid injection of alcohol in 8 patients who demonstrated extreme spasticity complicating paraplegia and to present 1 of these cases briefly. These results substantiate the value of this procedure originally reported by Shelden and Bors.

The necessity of relieving the extreme spasticity and mass reflex that complicate many cases of paraplegia is clearly evident. The pain accompanying these spasms is often severe, and interferes with the nourishment and nursing care of the patient. Decubitus ulcers are more prone to develop in patients with flexion contractures of the lower extremities, and treatment of these is rendered extremely difficult in the face of spasticity, or the mass reflex with its accompanying involuntary micturition. Rehabilitation to the stage of ambulation with walking calipers is impossible in the presence of extreme spasticity of the lower extremities. Munro[9] and others[5] have performed anterior rhizotomy for the relief of extreme spasticity in paraplegia. The section of posterior nerve roots to achieve this end has also been de-

* Fellow in Neurosurgery, Mayo Foundation, Rochester, Minnesota.
† Department of Neurology and Neurosurgery, New York Medical College, New York, N. Y.

scribed.[6] However, there are certain drawbacks to these procedures: (1) The paraplegic patient is often too extremely debilitated to withstand the operative procedure. (2) It is a permanent mutilating operation. James and Braden[8] have reported the successful use of curare in the treatment of muscle spasticity in paraplegia. However, in our hands, this drug has failed to alleviate spasm to any appreciable degree.[4] Six of the 8 patients reported in this series had received curare without any substantial beneficial effect.

Our series of 8 injections was carried out between March 1946 and November 1946, approximately coinciding with those of Shelden and Bors. The flaccidity produced in our cases lasted a minimum of 6 months. During this period of alleviation of the spasticity, treatment of the decubitus ulcers, steps towards the development of an automatic bladder, and physical rehabilitation can make very gratifying strides. In none of our cases did the spasticity return to the degree of severity present prior to the intrathecal alcohol injection. However, in 1 case it did return to a degree necessitating further therapeutic measures. One patient succumbed to intercurrent urinary infection. The remaining 6 patients have been followed from 12 to 18 months without recurrence of spasticity. In each of these cases the urinary bladder was extremely hypertonic prior to the injection, so that the development of a satisfactory "automatic bladder" was impossible as long as the spasticity and mass reflexes persisted. Following intrathecal injection of alcohol the urinary bladder became hypotonic or tabetic in type, but within the ensuing 3 to 6 months in each case regained sufficient tonus to expel urine automatically at periodic intervals. The treatment was used in 8 patients, in 3 of whom it was considered to have been a life-saving measure, due to the extreme debilitating condition to which the spasticity and mass reflex had contributed. Many physicians postpone a mutilating procedure such as anterior rhizotomy until the patient is no longer strong enough to withstand the operation. This operation, therefore, although a valuable one in many instances, cannot be used in all cases of extreme spasticity in paraplegia. The intrathecal injection of alcohol produces flaccidity immediately, with little or no permanent additional loss of neurological elements, and does not insult the already precarious general physical condition of many of these patients.

METHOD

The patient is placed on one side in bed, and the foot of the bed elevated 18 inches so that the alcohol will rise to the caudal section of the thecal sack. A No. 18 spinal needle is inserted intrathecally and 95 per cent alcohol is introduced slowly, not more than 1 cc. every 60 seconds. An assistant stands on the opposite side of the bed to check the patient's sensory level so that it is not allowed to rise above the level previously established. The assistant also gently attempts to straighten the lower extremities after each cc. of alcohol injected. When the lower extremities are flaccid and can be easily straightened, no additional alcohol is injected. Complete flaccidity

will result from 7 to 12 cc. of 95 per cent alcohol injected in this manner. The foot of the bed is kept elevated for 24 hours to allow fixation of the alcohol.

One case, which illustrates the dramatic effect achieved by the use of intrathecal alcohol, is herein briefly reported.

Case 8. W. K., a 29-year-old white male with an incomplete spinal cord lesion at the 8th thoracic segment, was subject to extreme flexor contractions and spasticity of the lower extremities and to the mass reflex. During these spasms the patient cried out and complained of excruciating pain, and demanded opiates for relief. He refused to take nourishment and declined in weight from 160 to 90 pounds. He acquired large decubitus ulcers over each greater trochanter and treatment of these in any manner was rendered ineffective by the intense spasm of the lower extremities. He became depressed and abusive, and very difficult to manage on the open ward. The urinary bladder dribbled continuously and a suprapubic catheter was inserted. Three months prior to the alcohol injection he refused any operative procedure. Alcohol injection was proposed to him when it was apparent that the patient would continue downhill to death if the spasm were not alleviated so that further treatment could be carried out.

Intrathecal injection of 8 cc. of 95 per cent alcohol resulted in immediate flaccidity of the lower extremities. Within 24 hours a complete change of personality was noted in the patient, who became cheerful and cooperative. Within 3 months skin grafting of the decubitus ulcers was carried out and the patient was up in a wheel chair. Six months following the injection the spasticity was still alleviated but the urinary bladder had regained considerable tonus, so that training of the bladder to achieve automaticity could be undertaken. In this case the intrathecal injection of alcohol was believed to be a life-saving measure.

SUMMARY

1. The use of the intrathecal injection of 95 per cent alcohol to alleviate spasticity in paraplegia is suggested in certain cases.

2. Its advantages over anterior rhizotomy in certain instances are cited.

3. The results of 8 cases in which this treatment was used are summarized and 1 case is presented in detail.

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EXTRADURAL CEREBELLAR HEMORRHAGE

REVIEW OF THE SUBJECT AND REPORT OF A CASE*

FRANK M. ANDERSON, M.D.

Neurosurgical Service, Children's Hospital, Los Angeles, California

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EXTRADURAL hemorrhage confined primarily to the posterior fossa of the skull is an unusual lesion of considerable clinical interest. Hemorrhage of this variety arises from a tear in some portion of the lateral sinus precipitated by head injury, but trauma need not be sufficiently severe to cause immediate loss of consciousness, and fracture of the skull may be absent. As blood accumulates in the extradural space overlying the cerebellum, signs of obstruction in cerebrospinal fluid outflow appear and evidences of cerebellar dysfunction become manifest. It is the purpose of this paper to review the clinical features of this syndrome and to present the case of a patient with such a hemorrhage who recovered following operation.

SURVEY OF LITERATURE

McKenzie[5] has recorded 20 cases of extradural hemorrhage, one of which was confined to the posterior fossa overlying the right cerebellar hemisphere. In another case the clot covered the left occipital lobe as well as the left cerebellum, and in a third patient clot was found to cover the right cerebellar hemisphere and extend upward over the posterior two-thirds of the right cerebrum. In the first case the patient died unoperated upon; in the second, only the posterior fossa clot was removed and the patient did not survive; in the third, the supratentorial hemorrhage was drained but posterior fossa clot was not detected, and this patient also succumbed.

Coleman and Thomson[1] reported 1 case of posterior fossa extradural hemorrhage in which the diagnosis was strongly suspected before operation. Three days after injury, cerebellar exploration was performed disclosing an extradural clot over the right cerebellum reaching to the left of the midline. The clot was evacuated and the patient recovered completely. Coleman[1] mentions a case seen by Mayfield in which posterior fossa hemorrhage was thought likely, but the patient died before operation could be performed. Extradural clot over the cerebellum was found at autopsy.

Turnbull[8] reviewed the subject of extradural posterior fossa hemorrhage and recorded a case of a woman who had sustained minor head injury with subsequent headache, then 9 months later a second injury, the latter precipitating drowsiness, blurring of vision, and unsteadiness of gait. Suboccipital craniotomy disclosed an extradural clot over the right cerebellar lobe. The clot was removed and the patient recovered except for mild visual impairment.

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Scoville,[7] Kessel,[3] and Watts[10] each described 1 case of such hemorrhage, and in these 3 instances operation was carried out, all the patients recovering. Watt's case is unusual in that the cerebellar operation was performed immediately after bilateral subtemporal exploration failed to disclose extradural hemorrhage over the cerebrum.

In a survey of 34 extradural hemorrhages Gurdjian and Webster[2] reported 1 case wherein the patient sustained a perforating wound of the head just above the lateral sinus, with formation of extradural clot over the cerebellum and occipital lobes. Evacuation of the clot resulted in prompt recovery. Munro and Maltby[6] appraised their 44 cases of extradural hemorrhage and recorded 1 instance of posterior fossa clot discovered at autopsy.

Autopsy material comprising 504 cranial injuries was presented by LeCount and Apfelbach,[4] who found 104 extradural hemorrhages of sufficient size to produce symptoms. Eight of these were associated with fractures of the posterior fossa, but it is not made clear whether the clot in these instances lay primarily over the cerebellum or over the occipital lobes. Vance[9] made an extensive appraisal of 512 skull fractures at autopsy and found sizable extradural hemorrhage in 61 cases. In 4 the clot was found over one or both occipital lobes as well as the cerebellum, and bleeding was found to have originated from a lateral sinus tear. In neither of the above autopsy studies is there record of clot confined to the posterior fossa.

REPORT OF CASE

J. F., a white girl 5½ years old, was admitted to Children's Hospital April 7, 1947. Twelve days previously she had fallen and struck the back of her head on a hard surface without losing consciousness. In the ensuing 4 days the child became lethargic and vomited frequently without nausea but did not complain of headache. During the following week she grew excitable, cried easily and tore her clothing, and at this stage the parents observed her eyes to be crossed.

Neurologic Examination. The patient was irritable, but lucid and fairly cooperative. Suboccipital tenderness was notable, more marked on the right side, and there was mild resistance of the neck to flexion. Bilateral papilledema, large pupils reacting poorly to light and convergence, and bilateral 6th nerve pareses were observed but nystagmus was not present. Although gag reflexes were normal the voice had a nasal quality and the soft palate was depressed, more so on the left than right. All extremities were moderately hypotonic and ataxic, the right arm and leg appearing slightly more unsteady than the left, and as the child walked she deviated fairly consistently to the right, holding her head inclined to this side. The deep reflexes were symmetrical and of small volume throughout; a Babinski sign was present on the left.

X-rays of the skull showed questionable suture separation but no fracture. Spinal puncture was not done. EEG tracings demonstrated phase reversal in the left occiput and were interpreted as suggestive of a deep-seated lesion below the left occipital lobe. On clinical grounds, intracerebellar hemorrhage and posterior fossa subdural hematoma were considered strongly in the diagnostic possibilities, but right cerebellar neoplasm was felt to be more probable than either of these.

April 10, ventriculography was performed through posterior parietal burr holes, 40 cc. of CSF being removed and replaced with an equal amount of air. This study showed moderate internal hydrocephalus, and forward dislocation of the 4th ventricle with narrowing in its anteroposterior dimension (Fig. 1).

FIG. 1. Ventriculogram (retouched) after instillation of 40 cc. of air, some of which has escaped into the subarachnoid pathways. Moderate dilatation of the lateral ventricle is apparent, and there is distinct dislocation forward of the cerebral aqueduct and 4th ventricle with anteroposterior narrowing of the latter structure.

Operation. Suboccipital exploration under endotracheal ether anesthesia was carried out immediately, using a curved transverse incision along the superior nuchal line. When the left occipital plate was perforated, dark liquid and clotted blood spurted through the opening under moderate pressure. The bony opening was enlarged and the posterior rim of the foramen magnum removed, disclosing the clot to be extradural, about 6 cm. in diameter and 1.5 cm. in greatest thickness, reaching from the midline to the left sigmoid sinus (Fig. 2, insert). There was no bleeding after evacuation of the clot, which came away readily from the dura. A small opening was made in the dura to exclude subdural clot and the left cerebellar hemisphere was needled once to be certain no intracerebellar hemorrhage was present. The cisterna magna was opened and the cerebellar tonsils were elevated gently from their position of impaction in the foramen magnum. No fracture of the skull was visualized and no other pathology was in evidence. The wound was closed in layers without drainage.

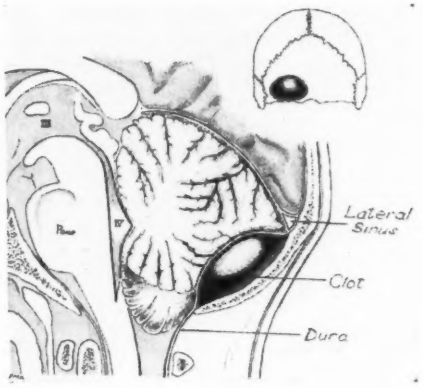

FIG. 2. Diagram illustrating relation of an extradural clot to structures in the posterior fossa. Distortion of the cerebellum, forward dislocation of the brain stem with consequent displacement of 4th ventricle and aqueduct, and compression of the 4th ventricle are indicated. Insert shows approximate size and location of hemorrhage in the case presented.

Course. The patient recovered rapidly. After 5 days the left abducens palsy was less marked, and a week later this defect had almost cleared, though paresis of the right abducens nerve was still prominent. At this time ataxia of the extremities was becoming less pronounced. Patient was discharged April 28, 1947. When seen in the outpatient department 2 months later, she was entirely free of symptoms, the only finding being small excursion nystagmus in left lateral gaze. The optic fundi were negative, the gait was normal and all extremities were used actively, without ataxia. Examination 5 months after operation showed almost imperceptible nystagmus in left lateral gaze, but no other findings. The child ran and turned quickly without impairment of any sort. She has remained well to the date of writing (1 year).

DISCUSSION OF CLINICAL FEATURES

Extradural posterior fossa hemorrhage is undoubtedly an unusual lesion, yet if one includes cases in which the clot reaches upward over one or both occipital lobes of the cerebrum, the incidence is not extraordinarily low. In 3 of McKenzie's 20 cases,[5] hemorrhage extended into the posterior fossa and, in 1 of these, it was confined entirely to the latter area. In Vance's material[9] the incidence of such clots is more than 6 per cent of the total extradural hemorrhages (4 out of 61), and LeCount[4] reported 8 clots involving the posterior fossa as well as the posterior portion of the supratentorial space, an incidence of nearly 8 per cent. At the Los Angeles County Hospital, no example of posterior fossa extradural hemorrhage has been noted in the past 14 years, although approximately 2,000 head injuries are treated each year and an average of 6 cases of epidural hemorrhage over the cerebrum are operated upon per year. No instance of extracerebellar hemorrhage has been recorded by the Los Angeles County Coroner in the past 12 years but precise records are not available and it is possible that posterior fossa clot has been overlooked in certain of these autopsies.

The case here presented is the only one of this variety recorded at Children's Hospital, but extradural hemorrhage in children and infants is notoriously rare and at this hospital only 2 extradural clots of consequential size have been found at surgery or autopsy in the last 15 years.

The clinical picture produced by extradural hemorrhage over the cerebellum has been well described by Coleman and Thomson[1] and the reported cases in which complete data are given show many of the criteria set forth by these writers. Their patient was a 9-year-old child who sustained injury to the back of the head but did not lose consciousness; 24 hours later he complained of headache, vomited twice, and became drowsy, although no significant neurologic findings were evident. Symptoms persisted, but not until almost 3 days after injury were objective neurologic abnormalities detected, consisting of stiffness of the neck, marked hypotonia, and a few hours later, loss of the deep reflexes; neither papilledema nor nystagmus was present. The child lay on his left side most of the time and resented being turned. X-Ray of the skull was normal except for linear fracture of the occipital bone, entering the foramen magnum. Operation on the 3rd day disclosed an extradural hemorrhage covering the entire posterior surface of the

right cerebellum as well as a portion of the left cerebellar hemisphere. The clot was removed and the patient recovered uneventfully.

Two of McKenzie's patients presented a clinical picture similar to that given above and McKenzie[5] makes careful note of the fact that flaccidity of the extremities was a particularly striking finding in his cases.

The case reported by Turnbull[8] is unique in that minor head trauma was followed for 9 months by intermittent headache, a symptom which suggests that posterior fossa clot may have formed after this injury. A second injury (9 months after the first) precipitated rather abrupt symptoms of drowsiness, increasing headache, slight unsteadiness of gait, and blurring of vision in the left eye. Two weeks later the patient had stiffness of the neck, bilateral choked discs, slight incoordination of the right extremities with diminished reflexes in the right lower extremity, and a tendency to stagger to the left. Cerebellar tumor was suspected and ventriculography was done, followed by suboccipital craniotomy 3 weeks after the second injury. Extradural clot was found over the right cerebellar hemisphere, but no comment is made regarding encapsulation or direct evidence that the clot was of several months' duration. Removal of the hemorrhage was carried out and the patient recovered fully except for mild diminution in visual acuity due to secondary optic atrophy.

The following data regarding symptomatology and findings in the presence of posterior fossa extradural hemorrhage are a composite of those brought out by previous authors, principally Coleman and Thomson,[1] and by appraisal of the case herein reported.

The patient is struck on the back of the head and may or may not lose consciousness; headache ensues, often accompanied by vomiting, and a lethargic, irritable state follows. After a period varying from a few hours to many days the lethargy deepens into coma. The patient may prefer to lie on one side (presumably the side of the lesion) and will resume this position if turned. Neurologic findings are often meagre, but neck rigidity, papilledema, nystagmus, ataxia, hypotonia, reduction or loss of deep reflexes, and tonic convulsions have been described at some time during the course of various cases, if the patient's cooperation has allowed adequate examination. Some of these signs are therefore to be expected, particularly the finding of flaccidity of the extremities. Because the hemorrhage is venous, arising from a tear in the lateral sinus, the clot often accumulates slowly and allows the "lucid interval" after injury to extend for several days (or months?),[8] during which time the patient may complain little and exhibit no abnormalities. More commonly, symptoms or signs of rising intracranial pressure have been noted in 24 hours or less, and it is clear that when coma supervenes in the untreated case there is rapid downhill progression with cardiac and respiratory irregularities, and death.

X-Rays of the skull may show fracture line in the occipital bone crossing the lateral sinus. In the presence of supporting clinical signs such a fracture is suggestive evidence in favor of extradural hemorrhage, inasmuch as tear-

ing of the lateral sinus may occur when the skull is fractured. Skull fracture need not be present, however. Spinal puncture has not given significant information and is probably best avoided if the diagnosis is reasonably certain.

Treatment of extradural posterior fossa hemorrhage consists in suboccipital craniotomy with removal of the clot and institution of appropriate repair measures if bleeding from the sinus is encountered. Drainage for 24 hours is advisable unless absolute hemostasis has been obtained. As in the presence of extradural hemorrhage over the cerebrum, operation is indicated early, before the advent of excessive intracranial pressure with accompanying coma and medullary decompensation. In most of the reported cases there was rather slow progression of symptoms enabling the examiners to observe and recheck the odd clinical problem presented and then to proceed as with cerebellar tumor. In the presence of more rapid hemorrhage, fulminating signs of cerebellar and brain stem compression will not permit extended observation, and in these patients prompt diagnosis and operation are imperative. It seems probable that the acute type of posterior fossa hemorrhage is, on occasion, solely responsible for death following head injury, and such deaths might by greater cognizance of this lesion be prevented.

SUMMARY

The subject of traumatic extradural hemorrhage over the cerebellum is reviewed and a case of this syndrome presented in which operation was performed, with recovery of the patient.

Hemorrhage in this location may cause acute compression of the cerebellum and 4th ventricle with abrupt rise in intracranial pressure, tonic convulsions, flaccidity of the extremities and rapidly deepening stupor. However, in several reported cases there were no remarkable symptoms for 24 hours or longer, when headache, lethargy, and "cerebellar" signs appeared and gradually increased in prominence.

As soon as hemorrhage of this variety is suspected, suboccipital craniotomy and evacuation of the clot should be performed.

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UNILATERAL HYDROCEPHALUS RESULTING FROM OCCLUSION OF FORAMEN OF MONRO

COMPLICATION OF RADICAL REMOVAL OF BRAIN ABSCESS

EBEN ALEXANDER, JR., M.D.,* AND E. HARRY BOTTERELL, M.D.
Department of Neurosurgery, Toronto General Hospital, and Department of Surgery, University of Toronto, Ontario, Canada

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THE treatment of brain abscesses in all stages of encapsulation by radical removal of abscess and adjacent softened brain tissue has been reported by several authors.[6,11,14,15] The reduction in over-all mortality using this method in conjunction with systemic and intraventricular penicillin is a consistent finding in all the reports in the literature. Chorobski and Kunicki[6], in a recent publication reviewing all reported brain abscesses treated by radical removal, state that there were no cases in which meningitis was reactivated by the operation provided antibodies were used.

In view of the growing enthusiasm for this type of treatment of brain abscesses, 2 cases are presented with unilateral hydrocephalus secondary to obstruction of the foramen of Monro following removal of a brain abscess.

The causal relationship between obstruction in the cerebrospinal fluid circulation and the development of hydrocephalus has been suspected for over half a century and proved experimentally since 1913. The first experimental production of hydrocephalus in animals by occlusion of one foramen of Monro by Thomas[20] was overshadowed by the publications of Dandy[9] at about the same time. Both workers gave adequate experimental proof of the development of hydrocephalus in animals following occlusion of the cerebrospinal fluid pathways.

Severe reactions in the ependyma of the lateral ventricles, 3rd ventricle, or aqueduct of Sylvius producing subsequent gliosis and obstruction to the interventricular passages have been occasionally reported. The cause of the ependymitis in some cases has not been determined.[2,12,18,19,21] In others the gliosis of the ependyma has been secondary to known or suspected meningitis.[1] Burr and McCarthy[4] as early as 1900 attempted to duplicate this picture by injecting irritating substances into the lateral ventricles of experimental animals. They successfully produced ependymitis but unfortunately did not allow their experimental subjects to survive long enough to develop dilatation of the ventricles.

The first clearly defined clinical case of unilateral hydrocephalus from obstruction to one foramen of Monro was described by Cushing.[8] In this presentation Cushing considered the obstruction was secondary to ependymitis, the result of so-called "brain fever" some months previously. The

* Bowman Gray School of Medicine, Winston-Salem 7, North Carolina.

number of cases reported in the literature is small and the surgical attempts to alleviate this condition have been remarkably few. Dott[10] in 1927 described a patient 9 months of age with obstruction of one foramen of Monro and severe unilateral hydrocephalus. The patient was apparently permanently relieved of symptoms of increased intracranial pressure by an opening, made at operation, through the *septum pellucidum*.

More recently Cairns *et al.*[5] have described 2 cases resulting from gunshot wounds of the head in which portions of the lateral ventricles had been sealed off from the rest of the ventricular system by scar tissue. These patients suffered from local dilatation of the lateral ventricle confined to its occluded portion of sufficient severity to cause generalized increase in intracranial pressure. In one patient communication between the occluded inferior horn of the lateral ventricle and the body of the ventricle was established and the choroid plexus within the cyst removed. In the other case the choroid plexus was removed. In both patients the increased intracranial pressure was relieved.

During the past 2 years one of us (E.H.B.) has treated 8 patients with brain abscess by means of radical excision. There has been 1 death. In 2 instances in which the abscess was poorly encapsulated with but thin walls and in close proximity to the lateral ventricle, the abscess was ruptured during removal and the adjacent ventricle opened. In both patients penicillin was used locally in the cerebral cavity and in one streptomycin was also used. Intraventricular injections of these antibiotics were also made postoperatively. The prompt recovery with complete bacteriological cure was impressive. The subsequent insidious development of signs which were mistakenly interpreted as recurrence of an abscess and satisfactory surgical cure of these patients when the true nature of the condition was found by ventriculography are related below.

PRESENTATION OF CASES

Case 1. B98276. R. P., male, 19 years.

Summary. Frontal osteomyelitis and left frontal lobe abscess. Radical removal of abscess; ventricle opened; intraventricular penicillin. Temporary improvement followed by gradual onset of signs of space-occupying lesion of left cerebral cortex. Unilateral hydrocephalus; obstruction left foramen of Monro, relieved by opening made through *septum pellucidum*.

Present Illness. This 19-year-old boy was in good health until Jan. 3, 1947, when acute left frontal sinusitis developed. On Jan. 9, 1947, a swelling in the left frontal region was drained and purulent material from this cultured *Streptococcus haemolyticus*. On sulfonamide therapy he made a gradual recovery until about Feb. 15, 1947, when he was admitted to the Neurosurgical Service of the Toronto General Hospital in semi-coma.

Examination. Temperature 101° F., pulse 60, respirations 10. He was drowsy, rather poorly nourished and responded, answering simple questions correctly. Findings were: edema of the left forehead, moderate bilateral papilledema with small retinal hemorrhages, and weakness of the right lateral rectus muscle. Power

in the extremities and superficial and deep reflexes were considered normal. There was no aphasia. The patient was right-handed.

Laboratory Data. Wbc. 9,200. Roentgenograms of the skull showed frontal sinusitis and osteomyelitis of the frontal bone (Fig. 1).

Shortly following admission the patient had a generalized seizure with respiratory arrest of alarming duration. Immediate operation was necessary.

Operation, Feb. 15, 1947. Using a left frontal scalp flap, the infected bone of the left frontal region was removed together with a safe margin of normal bone surrounding it. The left frontal sinus was obliterated. The dura was infected and there was a large abscess of the left frontal lobe. This was radically removed together with surrounding softened edematous brain and the anterior horn of the left lateral ventricle was opened. A burr hole was made in the right posterior parietal region and penicillin (5000 oxford u/cc.) inserted into the right lateral ventricle. Penicillin in the same concentration was used to irrigate the left frontal wound. The wound was closed tightly.

FIG. 1. Case 1. P-A film of skull showing osteomyelitis of left frontal bone and frontal sinusitis. Feb. 15, 1948.

Postoperative Course. He was given systemic penicillin and sulfadiazine, as well as local instillation of penicillin, 10,000 units, into the operative site on 2 occasions and 10,000 units of penicillin by lumbar route on 4 successive days after operation. He left the hospital much improved.

For 1 month after leaving hospital he gradually became increasingly drowsy, at times irrational and confused, and spent most of the time in bed.

He was readmitted on May 13, 1947. The frontal decompression on the left side was full; there was a right lower facial weakness as well as a right homonymous hemianopsia. The right arm and leg were weak, with hyperactive reflexes of the right leg but no pathological reflexes.

Reexploration of his left frontal region was carried out. There was no abscess and the left frontal horn of the ventricle was encountered only a short distance under the cortex. A ventriculogram was done 3 weeks later, June 4, 1947. This showed a greatly enlarged left lateral ventricle and a localized dilatation of the left frontal region, which it was felt might not have adequate communication with the rest of the ventricular system.

The patient showed improvement following this procedure and was able to be out of bed alone. By Nov. 3, 1947, he had gained some strength in his right side and was more active. Dysphasia was present at this time, which had not been noted previously. He was given intensive physiotherapy and occupational therapy, without improvement.

Readmission, Jan. 28, 1948. The patient had shown little change in his condition

until about 1 week before admission. He then began to be drowsy and to complain of severe headache. There had been an increase in the weakness of his right side. The dysphasia was more severe than previously. The scalp over the left frontal bone defect was tense and bulging.

It was again considered that there was a recurrence of his left frontal lobe abscess. Tapping the left frontal region through the defect, however, did not reveal an abscess but rather the anterior horn of the left lateral ventricle, containing clear colorless fluid under pressure.

Ventriculography, Feb. 4, 1948 (Figs. 2 and 3). Air was first inserted in the right

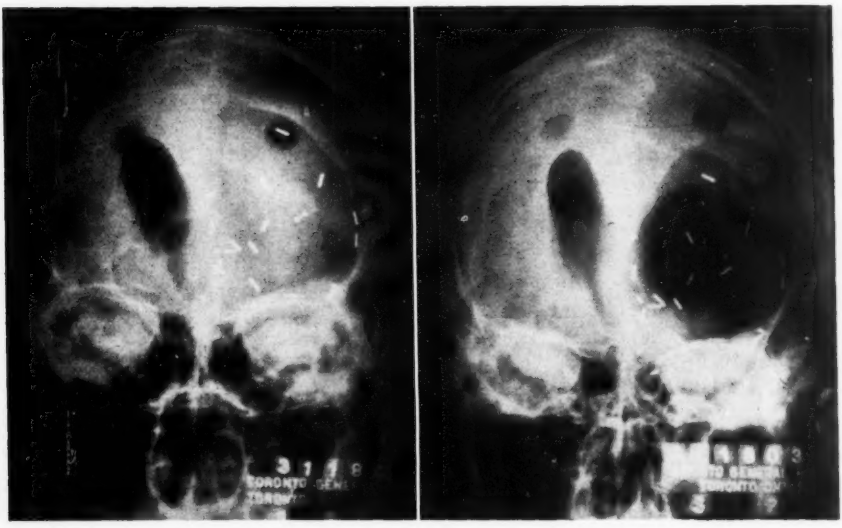

FIG. 2 (*left*). Case 1. A-P film of skull after air injected into right lateral ventricle. Shift of 3rd ventricle to the right. No air has passed to the left side. Feb. 4, 1948.

FIG. 3 (*right*). Case 1. A-P film. Ventriculogram. Air injected directly into both lateral ventricles demonstrating gross dilatation of anterior portion of left lateral ventricle and shift of *septum pellucidum* to right. Feb. 4, 1948.

occipital horn. Air was seen in the right lateral ventricle, which was shifted slightly to the right. There was no air in the left side. The left frontal horn was tapped and air introduced into this ventricle. A large cystic cavity was seen.

Operation, Feb. 5, 1948. The left frontal region was reexplored. There was considerable scarring of the frontal lobe. Exploration of the left lateral ventricle showed extensive scarring about the site of the foramen of Monro and no opening could be made out. The remainder of the visible ependyma did not appear abnormal. An opening was made in the *septum pellucidum* establishing communication with the right lateral ventricle. Since air had been previously injected into the right lateral ventricle there was no free flow of CSF between the ventricles but it was felt that a definite communication had been established.

Postoperative Course. The patient was much brighter and showed an improve-

ment in the function of his right arm and leg. He was mentally much more alert and his dysphasia began to decrease. Although still severely disabled, the improvement was striking and he was discharged.

When carefully examined 6 months after operation he showed further improvement. He came 40 miles to the clinic, alone, by bus. He has had no convulsions or headaches. He walks with a hemiplegic gait but can hold objects in his right hand. The right facial weakness is barely perceptible. The dysphasia is evident only on careful testing and he can carry on an intelligent conversation without noticeable difficulty. The homonymous hemianopsia persists and is complete. Optic discs and visual acuity are normal.

Comment. The intermittent onset of symptoms in this patient with periods of apparent improvement led to a mistaken interpretation of the underlying cause of his signs and symptoms. Ventriculography done 9 months prior to final operation might have led to a correct diagnosis if air had been injected into one ventricle only. An intermittent incomplete block would have explained the patient's picture, and ventriculography performed by injecting air into one side only might not have given conclusive information at that time.

In spite of the fact that the second patient entered the hospital for his last admission only a few weeks after the dramatic improvement of Case 1 following operation, the true nature of the condition was not suspected until ventriculography was done. Once the condition was shown in its proper light, certain details learned from Case 1 were used to advantage.

Case 2. B25262. H. S., male, 30 years.

Summary. Right occipital abscess secondary to bronchiectasis. Radical removal of abscess; exudate in ventricle; intraventricular antibiotics. Temporary improvement followed by gradual onset of signs of space-occupying lesion in right cerebral hemisphere. Obstruction right foramen of Monro; unilateral hydrocephalus relieved by opening in *septum pellucidum.*

History. In 1934 this patient had bronchopneumonia; bilateral bronchiectasis followed this, proved by lipiodol bronchograms. The most severely involved portions, the left lower lobe and the lingual process of the left upper lobe, were removed by Dr. Robert Janes on Oct. 8, 1942. Sputum cultured *Streptococcus haemolyticus.* Marked improvement followed this operation. During the succeeding 4 years the patient raised only a small amount of sputum, and was able to work steadily in a cold climate without any recurrence of pulmonary infection.

Present Illness. On Sept. 1, 1947 there suddenly developed a severe right frontal headache which persisted almost without remission. He had shown no signs of recurrent pulmonary infection, and had continued to expectorate only small amounts of sputum. Examination by his family physician showed no cause for headaches but a spinal puncture revealed normal pressure, with increased globulin, 250 lymphocytes per mm.; and a first zone rise in the gold sol curve. He was admitted to Toronto General Hospital, Sept. 15, 1947.

Examination. He was found to have a left homonymous hemianopsia with macular sparing, no papilledema and no evident weakness of the left side.

Operation 1, Sept. 22, 1947. Ventriculography was done. This demonstrated a space-occupying lesion of the right occipital region which at subsequent operation

proved to be a very large abscess (Fig. 4). There was little or no capsule; the abscess, together with a large area of surrounding edema and encephalitis in the occipital and parietal lobes, was removed. In the process of removing the abscess it was ruptured. The tentorium was exposed and the ventricle opened; there was evidence at this time of exudate over the choroid plexus, and of cloudy ventricular fluid. The patient was given systemic penicillin and sulfadiazine; 25,000 units of penicillin (5000 u/cc.) were injected into the *right* lateral ventricle and cerebral defect. The wound was closed tightly and the bone flap sacrificed.

Three hours after operation the frontal horn of the right lateral ventricle was tapped and 100,000 units of penicillin and 1,000,000 units of streptomycin (1 gm.)

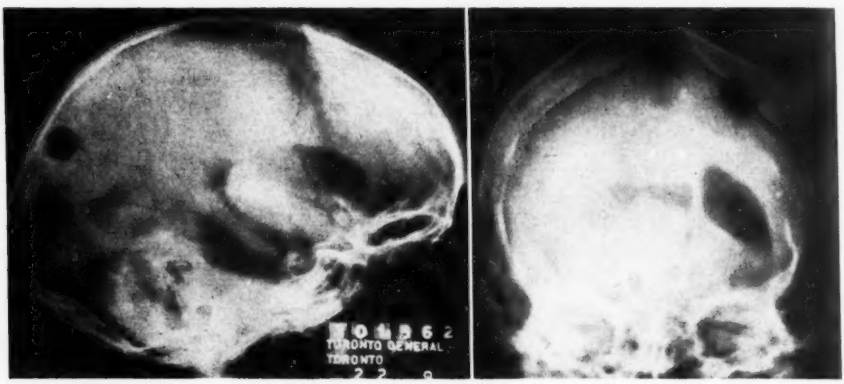

FIG. 4. Case 2. Left lateral and P-A films of skull. Ventriculogram demonstrating right occipital lobe mass lesion. This was a large abscess of the occipital lobe. Sept. 22, 1947.

were injected into the ventricle. The solutions containing these drugs were intended to be of substantially lower concentrations, but due to the error of an assistant in preparing them, the above were inadvertently used. The patient had a sharp rise in temperature to 104°F. the evening of the operation, but the following day he showed improvement and from that time until his discharge from the hospital his condition was satisfactory. Moderate concentrations (25,000 units) of penicillin and streptomycin were given intraventricularly through a right frontal burr hole for 3 days and continued for 4 days by the lumbar route. Systemic penicillin and streptomycin were continued for 6 days, sulfadiazine having been discontinued on the 2nd postoperative day because of hematuria. There was no infection of the wound and the patient left the hospital apparently free of headaches but with a persistent left homonymous hemianopsia; there was no hemiparesis. Culture of the pus from the abscess showed no organisms.

For 2 weeks following discharge, the patient was well; then he began to show personality changes. He talked irrationally, was dependent on his wife, and became incontinent of urine and feces. Left hemiplegia gradually developed, which was at some times more severe than others. There was no significant increase in pulmonary symptoms.

Readmission, Feb. 7, 1948. The patient was euphoric, disoriented, but cooperative. He showed evidence of dehydration. His attention could not be held in any

conversation. Neurological examination revealed fullness of the parieto-occipital bone defect, left homonymous hemianopsia, no papilledema, left lower facial weakness, weakness of both the left arm and left leg, incontinence of urine and feces. He was right-handed.

It was considered that he had a recurrence of his brain abscess or a second abscess located in the intracranial cavity. Sulfadiazine and penicillin were started systemically and intravenous fluids were given. He showed slight improvement but in general over the next month his condition remained much as on admission. He was listless, remained in bed, showed a persistent left hemiparesis, and was incontinent of urine and feces.

Operation 2, Mar. 8, 1948. Ventriculography. With the head turned on the right side, needles were inserted into both lateral ventricles through the posterior parietal burr holes. Clear colorless fluid dripped from both needles and the fluid was replaced with air in 10 cc. increments through the right needle only. A small amount of air was thought to have passed from the right to the left ventricle but only a small fraction of that injected. Air was injected into both ventricles before the patient was sent to the Roentgenology Department. The roentgenograms (Fig. 5) showed shift of the *septum pellucidum* to the left side with an enlargement of the right lateral ventricle. No air could be induced to enter the 3rd ventricle.

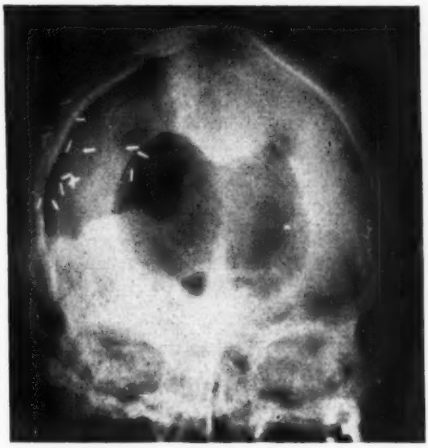

FIG. 5. Case 2. A-P film of skull 6 months after removal of abscess. Air injected into both ventricles. Shift of *septum pellucidum* to left. Greatly dilated right ventricle. Superimposed right occipital bone defect. Mar. 8, 1948.

Under local anesthesia a right frontal bone flap was turned down. A burr hole was likewise made in the left frontal region and the tip of a ventricular needle passed into the anterior horn of the left ventricle. Through a cortical incision the right lateral ventricle was entered and thoroughly examined. The site of the foramen of Monro was easily made out but there was no visible opening. The ependyma did not appear grossly abnormal. The ventricular needle in the left anterior horn could be seen against the *septum pellucidum* and, using it as a guide, a large opening was made in the *septum* directly into the left ventricle. Isotonic saline was washed through the opening repeatedly using the needle in the left lateral ventricle, demonstrating the patency and size of the new ostium.

Postoperative Course. The patient showed immediate and dramatic improvement. His mental state cleared, he became continent of urine within 3 days and was out of bed of his own accord on the 5th postoperative day. The left hemiparesis cleared almost completely except for a slight lag in the left lower face on emotional expression. The right parieto-occipital bone defect was depressed and there was now no evidence of increased intracranial pressure. He continued to show daily improvement during the rest of his hospital stay and was discharged mentally clear and anxious to return to work. An X-ray of the chest shortly before discharge showed clearing as compared to previous films. There was still some clouding of the left base.

On April 1, 1949, the patient reported he was working full time as a clerk, taking a course in accounting, free of headaches and unaware of any weakness of the left arm or leg.

Comment. The apparent passage of a small amount of air from the right to the left lateral ventricle may have been due to the existence of a minute opening at the foramen of Monro not visible at operation. The great enlargement of the right lateral ventricle with the shift of the *septum pellucidum* to the opposite side would lead to the conclusion that there was a serious obstruction to the flow of cerebrospinal fluid through the right interventricular foramen. Since in this case, as in Case 1, the fluid in the enlarged ventricle was clear and colorless, it is probable that the occlusion of the foramen of Monro was not complete but of such importance as to cause increased intraventricular pressure.

The clinical improvement in this patient following the removal of the left lower lobe of his lung, in spite of the fact that other lobes were involved in the bronchiectatic process, is of passing interest. This improvement has now persisted for 6 years.

The fact that the first brain abscess was metastatic in origin led those who observed the patient during his last admission to feel that his symptoms were due to a second abscess of similar origin. In view of the well known tendency of metastatic brain abscess to be multiple,[7,17] the solitary nature of the abscess in this patient is worthy of comment.

Although in cases with enlargement of both lateral ventricles and without scarring of the ventricular walls it is a simple matter to identify and open the *septum pellucidum*, it may be difficult to be certain of landmarks in cases such as those presented in this report.[5] The procedure of placing a ventricular needle in the normal ventricle at the time of operation, as was done to advantage in Case 2, may be of assistance in the surgical treatment of similar cases in the future. The location, patency, and size of the artificial opening of the *septum pellucidum* can be readily demonstrated by this procedure.

DISCUSSION

The type of operation carried out in these cases was so direct and simple and the therapeutic results so satisfactory that it would seem advisable to avoid in future cases the long delay brought about by the mistaken impression that a second or recurrent abscess was the cause of the signs and symptoms.

There was, in each case, a dramatic recovery from the removal of the brain abscess, a period of improvement which lasted 1 to 3 months. Following this there was recurrence of headache, mental and physical lethargy, with subsequent development of contralateral hemiparesis. That hemiparesis can be caused by unilateral hydrocephalus has been previously recorded.[5,8,10] In Case 1, in which the hydrocephalus occurred on the left side, the patient

also showed a partial aphasia. The course in each case was progressively downhill, though interrupted by periods of seeming improvement in Case 1, and before the final admission to the hospital each was bed-ridden, one of them incontinent of urine and feces.

The observation, at the time of removal of the brain abscess, of exudate over the choroid plexus and cloudy cerebrospinal fluid seems sufficient cause, in itself, for the later development of obliterative ependymitis and gliosis. Such late complications of bacterial infection of the central nervous system even after apparent bacteriological cure with antibiotics have been graphically illustrated by Bailey[3] and by Alexander.[1] Whether the intraventricular instillation of the antibiotic, penicillin, in one case with the addition of streptomycin in the other patient, both in relatively concentrated form, had any part in the ependymal reaction, it is difficult to say.[13,22] Certainly, this might have been a factor in Case 2 in which inadvertently a greater concentration than was desired was put into the ventricle. Nevertheless in patients with direct invasion of the ventricles with organisms, the local use of antibiotics is justified, for without this form of therapy, infection may not be controlled.[16,22] The frequent injection of solutions in low but effective concentrations would be advisable, avoiding concentrated solutions.[13]

A carefully performed ventriculogram should be done at an early stage in any patient who has been operated upon for a brain abscess but subsequently shows a persistent interruption in the course of improvement. This would determine the location of any recurrent abscess. Such studies, done earlier in the course of illness of the 2 patients presented herein, would have led to prompt diagnosis and appropriate treatment.

SUMMARY

1. Two cases of unilateral hydrocephalus from obstruction to the foramen of Monro have been presented.

2. Each patient had been successfully treated for a brain abscess by a radical removal of the abscess. In one patient the ventricle adjacent to the abscess was involved in the inflammatory process and in both patients the ventricle was opened at operation.

3. The relationship of the injection of concentrated solutions of antibiotics into the ventricles and the subsequent closure by scar tissue of the foramen of Monro has been discussed.

4. Attention is called to unilateral hydrocephalus as a cause of progressively deepening stupor with hemiparesis.

5. The relief of increased intracranial pressure by an artificial opening made surgically through the *septum pellucidum* was dramatic.

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FURTHER STUDIES ON THE TREATMENT OF EXPERIMENTAL HYDROCEPHALUS

ATTEMPTS TO DRAIN THE CEREBROSPINAL FLUID INTO THE PLEURAL CAVITY AND THE THORACIC DUCT

FRANC D. INGRAHAM, M.D., ROBERT A. SEARS, M.D., ROBERT P. WOODS, M.D., AND ORVILLE T. BAILEY, M.D.

Departments of Surgery and Pathology, Harvard Medical School, The Neurological Institute, and Department of Pathology, The Children's Hospital, Boston, Massachusetts

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As parts of a long-time study of experimental hydrocephalus in this laboratory, previous reports have described a reliable method for the production of obstructive hydrocephalus[4] and experiments with shunts into various venous channels.[5] The present paper deals with shunts of the cerebrospinal fluid into the pleural cavity and into the thoracic duct. We have felt that these procedures must receive thorough trial in experimental animals for their evaluation. Although our experiments have led to no new procedures for clinical use, they seem worth publication to prevent duplication in other laboratories and to take advantage of the opportunity to analyse the dynamics of cerebrospinal fluid when shunted outside the central nervous system and the causes of failure.

It has been assumed in these investigations that the cerebrospinal fluid arises from the choroid plexuses of the ventricular system, passes in a unidirectional current through the ventricles and gains access to the subarachnoid cisterns at the base of the brain by way of the laterally placed foramina of Luschka and the midline foramen of Magendie (held to be absent in dogs).[1] Thence the fluid passes into the adjacent subarachnoid cisterns, proceeding rostrally, and ultimately spreads out over the entire cortical substance. It is also free to pass down the central canal and the subarachnoid spaces of the spinal cord. Presumably some degree of absorption occurs in each subarachnoid recess, but the great bulk of the fluid is absorbed over the surfaces of the cerebral hemispheres and specifically by way of the arachnoid villi into the dural venous sinuses.

Experimentally, it is possible to produce hydrocephalus of several types. The ventricular pathways may be obstructed by the introduction of foreign objects or substances so that an internal hydrocephalus results. The aqueduct of Sylvius and the foramina of Luschka and Magendie are the strategic narrows most readily closed, but it is possible to occlude the interventricular foramen so that dilatation of one or both lateral ventricles results.[1] Arachnoidal sclerosing solutions have been introduced in such fashion as to seal the cisterns about the base of the brain, as by passing an iodine-soaked gauze completely around the brain stem,[1] with resultant dilatation of the

entire ventricular system and the proximal subarachnoid spaces. This is a true communicating hydrocephalus.

Clinically, as well as experimentally, attempts at surgical relief for either of these conditions have been directed either at the removal of varying portions of the choroid plexuses, or at by-passing the block. In the by-passing operations, the fluid is led either to the subarachnoid space beyond the block, or to a surface outside the nervous system which is capable of absorbing or draining the fluid. The present studies are confined to shunts designed to conduct the fluid to sites of drainage or absorption outside the nervous system. The relatively small brains of most experimental animals possess, even when advanced hydrocephalus is present, ventricles which on an absolute scale are far smaller than those dealt with in clinical practice. As a result, it is difficult to arrange a drainage tube within the ventricle so as to guarantee its continued function after anesthesia has ceased. The animals move violently, and the ventricle, having been more or less completely decompressed, is altered in size and shape.

Experimental Material. Dogs were rendered hydrocephalic by introducing a cellophane cylinder into the aqueduct of Sylvius mechanically through a suboccipital approach.[4] The occlusion was brought about slowly as a result of the irritative properties of the cellophane and, in the course of 1 to 3 weeks, led to moderate or marked dilatation of the ventricular system rostral to the point of occlusion (internal hydrocephalus). It seems probable that the reason that occlusion by a cellophane cylinder produces hydrocephalic animals more regularly than occlusion by a solid plug of foreign material is that cellophane leads to a slowly increasing obstruction to the flow of cerebrospinal fluid.

VENTRICULO-THORACOSTOMY

At its highest, the intrathoracic pressure in a normal animal may well be low enough to evacuate a distended ventricle so rapidly that proper compensation by the cerebral tissues cannot take place. The pleura, if it acts as an absorptive surface for cerebrospinal fluid, can take care of the amount that the choroid plexuses of the 3rd and lateral ventricles could be expected to produce.

In 2 dogs a polyethylene catheter, internal diameter 1 mm., was introduced into the left pleural space, its outer end heat-sealed and buried beneath the skin. Through this tube, 15–20 cc. of sterile normal saline were injected daily for 10 and 12 days, respectively. Neither dog had respiratory embarrassment at any time during these experiments. At autopsy there was no free fluid in the pleural space of either dog, and in both animals the lungs were fully expanded.

Operative Procedure. When one of the dogs previously subjected to occlusion of the cerebral aqueduct showed the characteristic signs of increased intracranial pressure,[4] a ventricular tap was carried out through the previously established posterior parietal burr hole. Anesthesia was used unless the animal was in coma. If the tap

indicated that the ventricle was enlarged sufficiently, as judged by the depth at which the needle entered it, preparations for the first-stage operation were made. The interval between insertion of cellophane in the aqueduct and operation varied between 1 and 3 weeks.

The first step was carried out under intravenous nembutal anesthesia with the dog prone, the head being slightly elevated in a headrest. A postparietal burr hole was made and enlarged with rongeurs to a diameter of 1.5 cm. Bonewax and absorbable hemostatic agents were used freely to ensure a perfectly dry dural surface. The dura was then perforated in the center of its exposed surface, and a lumbar puncture needle advanced until the occipital horn of the lateral ventricle was entered. This depth was recorded, and the needle withdrawn with the loss of as little fluid as possible. A section of zephiran-sterilized polyethylene tubing, 1 mm. in internal diameter, was inserted along the needle tract to this depth, and then advanced 2 mm. more. If satisfactory communication with the ventricle was demonstrated, the tube was grooved on its external surface with a hot needle at the level of the dura, and tied as securely as possible by means of sutures taken through the adjacent dura. The tube was considered functional only if the fluid within it was free to obey the laws governing fluids within a manometer. Satisfactory evidence of communication was generally obtained during anesthesia, and during the brief period required for completion of this portion of the procedure. The wound was closed in layers, the thick temporal muscle and its tough fascia being used as additional support for the ventriculostomy tube. Before closure of the skin, a subcutaneous tunnel was constructed by blunt and blind dissection as far down the neck as possible. The end of the tube was closed by crushing it in the jaws of a hemostat after heating. This sealed end was passed into the subcutaneous tunnel, and the skin closed above it.

Additional security was obtained for the ventriculostomy tube by the use of a flanged outer tubing of slightly larger diameter. The flange consisted of a circular piece of polyethylene film 0.02 cm. in thickness, which was attached by heat to the end of the tubing. This tubing was inserted first along the needle tract, since it had been cut to precisely the depth of the ventricle. Its flange thus came to lie on the dura and was cut to a slightly larger diameter than the bony defect. The overlapping edge was securely tucked between the dura and bone so as to prevent the tube from being driven too deeply into the brain. The arrangement was similar to the gold tube used by Miculwicz, as described by Davidoff.[2] The inner tube was then advanced through the flanged tube and tied securely at a depth 2 mm. greater than that recorded with the lumbar puncture needle. Some slight further support was gained by slipping over the inner tubing a second flanged tube, identical with the first, but previously curved by heating over a bent stylet so that when cooled it retained approximately the curve of the muscle which was to be closed around it. This second flanged tube was put in place with the flange inward, so that its flange abutted against that of the inner tube, and was also tucked between dura and bone. One such tube was demonstrated to be still in satisfactory communication with the ventricle after 13 days, at which time the animal was sacrificed.

Having obtained a functional ventriculostomy, thoracostomy was carried out through a short intercostal incision in the 2nd or 3rd interspace at the anterior axillary line on the side of the ventriculostomy. Dissection was carried down to the pleura and complete hemostasis secured. The pleura was opened with a small stab wound, and about 5 cm. of a second section of polyethylene tubing introduced into the pleural space. If the step was carried out rapidly, and pleural defect promptly

closed, pneumothorax was not a problem. If a more lengthy procedure was necessary, an endotracheal catheter and a positive pressure apparatus were used.

The tubing was fastened by means of silk sutures through the intercostal fascia, and further support gained from the muscle layers as these were closed tightly around the tube. The outer end of this tubing, hitherto sealed to prevent pneumothorax, was opened and connected with a water manometer, and pressure determinations made. Routinely, such pressures fell steadily from their initial height, showing a rhythmic swing with respiratory movements, until the final negative pressure of the thorax was recorded.

When the thoracostomy tube was functioning well, its outer end was again sealed, and passed subcutaneously as far toward the lower end of the ventriculostomy tube as possible. A third short incision was made over the palpable ends of both tubes in the neck, and these ends delivered into the wound. By means of a three-way stop-cock, these ends were then joined to each other and to a water manometer. Pressure determinations of ventricle and thorax were made. Next, the stop-cock was turned so that fluid flowed from the ventricle into the chest. Its progress could be watched through the transparent tubing, and frequent pressure determinations made. Results are presented in tabular form (Table 1). Two additional animals were excluded from the series because of operative infections.

Comment. Of the 23 total instances in which this system was observed to function well under the stable conditions of anesthesia, 16 ultimately failed later because of blockage. Table 1 shows that the chief cause of failure in these shunts was located in the ventriculostomy. The ventricular tube became blocked at least once in each of the 12 dogs in which this system was established. Three of the 6 ventriculostomies that were found to be functioning on re-exploration showed occlusion of the tubing distally to the ventricular end, so that the ventricle was not decompressed at the time of re-exploration. Of the 3 shunts that were actually functioning at the time of re-exploration, 1 was partly occluded at the ventricular end. When the tubing was occluded, this was usually due to a plug of cerebral tissue but fibrinous material was occasionally found.

The second most frequent site of obstruction was at the thoracic end of the shunt. Eleven of the 23 shunts showed occlusions of this portion of the tube, either as its sole apparent cause for failure, or in combination with other causes. Fibrinous plugs were commonest, but even here, cerebral tissue was sometimes encountered.

In 3 of the 23 shunts, separation of the anastomosis was the apparent cause for failure. This, of course, is a defect which can be avoided with improvements in the technic of joining the sections of tubing.

The features of the thoracic space which first made it appear favorable as a site for drainage have not been adequately tested by these experiments. In the two experiments cited, the chest was able to accept daily instillations of 10 to 15 cc. of saline without losing its negative pressure and without accumulating fluid. Whether or not this negative pressure is a desirable attribute in the drainage of cerebrospinal fluid is a debatable point. It may be

TABLE 1

Ventriculo-Thoracostomy

Dog No.	Re-exploration	Cause of Failure			Autopsy
		Ventriculostomy	Anastomosis	Thoracostomy	
N-33	5 days Functioning	O.K.	O.K.	O.K.	Chest free of fluid
	17 days No function	Occluded	O.K.	O.K.	
	—new shunt installed—				
	4 days No function	Occluded	O.K.	O.K.	Small subdural hematoma
N-112	5 days Functioning	O.K.	O.K.	O.K.	Chest clear
	15 days No function	Occluded	O.K.	O.K.	
	—new ventriculostomy—				
	3 days No function	Occluded	O.K.	O.K.	
N-116	2 days No function	Occluded	O.K.	Occluded	Chest clear Advanced hydrocephalus
N-192	2 days No function	Occluded	Separated	Occluded	Chest clear Only mild hydrocephalus
	—irrigated—				
	2 days No function	Occluded	O.K.	Occluded	
	—irrigated—				
	2 days No function	Occluded	O.K.	Occluded	
N-188	1 hr. No function	Occluded	O.K.	O.K.	Chest clear
N-163	1 hr. No function	Occluded	O.K.	Occluded	Chest clear
N-167	1 day Poor	Partly occluded	O.K.	O.K.	Chest clear Moderate hydrocephalus
	—ventriculostomy adjusted—				
	2 days No function	O.K.	O.K.	Occluded	
	—thorocostomy irrigated—				
	6 days No function	O.K.	O.K.	Occluded	
	—thoracostomy irrigated—				
	4 days No function	Occluded	Separated	Occluded	
N-128	6 days No function	Occluded	O.K.	Occluded	Chest clear
N-98	3 days No function	Occluded	O.K.	Occluded	Hemorrhage, cerebral cortex and basal ganglia
	—new ventriculostomy—				
	10 days No function	Occluded	Separated	O.K.	
N-67	3 days Functioning	O.K.	O.K.	O.K.	Pneumonia
N-64	7 days No function	Occluded	O.K.	O.K.	Entire shunt in chest No reaction
	—new ventriculostomy—				
	7 days No function	Occluded	O.K.	Occluded	
N-61	4 mins. No function				
—	—	—	—	—	
12	23	16	3	11	

that the negative pressure of the thorax does not account for the failure of the ventriculostomies, but the facts at least suggest this. The ventriculostomies that were found patent and in good communication, were found in those cases in which a block had occurred elsewhere in the system. In other words, a ventricle decompressed beyond a certain optimal point, becomes

a difficult space in which to keep a rigid tube patent. So long as the ventricle remained dilated, the ventriculostomy tube was usually in communication with it.

This situation might seem to exclude the hope of a successful shunt if it were not for the compromise offered by recipient sites in which the pressure never falls below a favorable minimum and never rises above a favorable maximum. Those shunts which have to date proved themselves theoretically and practically sound, such as the various internal ventriculostomies, including the Torkildsen operation, deliver the fluid into the subarachnoid space, which fulfills these conditions.

A further consideration is the relatively rigid quality of the polyethylene tubing used in these experiments. The soft ependymal wall of the ventricle is easily lacerated by even a slight movement of the tubing. Indeed, the debris which was found occluding the systems above, was usually found to be cerebral tissue. Further, many of the autopsy specimens showed corresponding gross defects in appropriate positions in the ventricular walls. The procedure used by Torkildsen[6] is routinely carried out in human subjects, using polyethylene tubing, or relatively stiff rubber catheters. However, the absolute size of the ventricular system in even the normal human brain is larger than that of the most advanced of the hydrocephalic animals used in this series.

From these studies it is clear that shunts between the hydrocephalic ventricles and the pleural space should not be attempted in human subjects with the technics that we have tested. Even though the difficulties with the central end of the tube are less in the larger ventricles of human hydrocephalic patients than they are in dogs, there remains the likelihood that the thoracic end may be sealed off by reaction of the serous surface.

VENTRICULAR-THORACIC DUCT SHUNTS

(Ventriculo-Lymphangiostomy)

Continuing the search for a favorable site into which to divert the cerebrospinal fluid, the advantages of a vessel, as opposed to an absorptive surface such as the pleural space, were recalled. The majority of the venous channels adaptable to this purpose have already been tested in this laboratory[5] and elsewhere, without success. This leaves for consideration only arteries and lymphatics. The high arterial pressure makes this system appear impractical, although review of the literature reveals no record of an attempt at utilizing these vessels in short-circuiting operations. Of the lymphatics, however, no obvious mechanical or physiological difficulty was apparent. The thoracic duct itself, being the largest and most constant lymph vessel, was selected as the recipient vessel. The general plan was to divert cerebrospinal fluid from an obstructed ventricular system into the terminal segment of the ligated thoracic duct.

It is a well established fact that the thoracic duct can be tied off completely and permanently in an otherwise healthy animal, with no untoward

results. Complete blockage of all the main lymphatic trunks is inevitably fatal. Ligation of the thoracic duct, alone, however, does not achieve complete lymphatic blockage.[3]

Preliminary studies were directed at investigating the competence of the valves guarding the orifice of the thoracic duct as it enters the venous system. In the 22 dogs examined, all possessed valves at this site which were competent as long as they were observed directly under conditions of surgical anesthesia. Even with the duct ligated, and with the venous pressure raised artificially, no reflux of blood into the duct was seen. This was in striking contrast to the situation found in the venous system, where incompetence of as many as three consecutive valves resulted when the venous pressure was raised.[5]

Studies carried out to determine the pressure within the thoracic duct indicate that it is extremely variable, ranging from 15 to 35 cm. of water, by end pressure determinations.[3] However, the pressure within the segment of duct proximal to the ligature is the pressure against which an obstructed ventricular system must be expected to compress itself. Presumably, the situation is entirely analogous to that found in isolated venous segments[5] where the pressure opposing the cerebrospinal fluid was the pressure within the vein into which a segment drained.

It was first considered desirable to cannulate the duct in the neck, thus making the by-passing tube as short as possible, and also avoiding the more hazardous procedures involved in opening the pleura extensively. Examination of this portion of the duct showed that the accessible portion of the duct was short, dangerously near the pleural dome, and variable in anatomy, being broken into two, three, and four separate channels, all too small to be cannulated satisfactorily.

Still hoping to avoid the chest, the abdominal lymphatic vessels were considered. In 3 dogs, these vessels were frequently found to be of adequate size, but were extremely friable, as was the cisterna chylae. Furthermore, use of these vessels involved certain entrance of chyle into the duct from tributaries entering proximally to the point of cannulation. Since lymph clots, this seemed an undesirable feature.

The duct, wherever it was approached in the thorax, was generally found to receive one or more collecting trunks higher in its course, which were ligated and divided. Cervical lymphatics almost always enter the duct well above the thoracic inlet, and these would escape. However, in no case was reflux of lymph into the transparent cannula observed either at operation or later. It proved helpful to give the animal a heavy fat meal the evening before operation. When this was done, the duct was large and contained milky chyle under pressures up to 40 cm. of water.

A second variable was the spasm of the duct in response to handling. Anesthetic ether gave satisfactory relaxation if applied before the duct was handled. Once spasm had occurred, however, attempts at cannulation were best delayed for 15 or 20 minutes, when relaxation returned.

Operative Procedure. The technique most successfully employed was a simple one. To assist in locating the thoracic duct, 5 to 10 cc. of Evans's blue dye were injected subcutaneously in a lower extremity. After brief, gentle massage of the injection site, the duct became readily visible as a brilliant blue streak within 20 minutes. The duct was dissected out of its bed just enough to allow a ligature to be passed beneath it. The ligature was left in place but not tied. The duct was grasped with fine smooth forceps, and a tiny incision made in its wall with a knife or with very sharp scissors. A section of polyethylene tubing, 1 mm. in internal diameter, was cut with a long bevel and passed gently into the duct for 1 cm. or more. The ligature was then tied securely around this. Additional security was obtained by first grooving the cannula about its circumference with a hot needle, and placing the ligature about this groove. The lower segment was then ligated. The loss of considerable quantities of lymph into the mediastinal space led to no apparent difficulties. Additional sutures were then taken through the periosteum of adjacent ribs, and tied around the cannula. The chest was closed tightly, the collapsed left lung being fully re-expanded. Care was taken to close the mediastinal pleura over the cannula, the outer end of which was then brought up through the wound. Pressure measurements of the fluid instilled into the cannula proved variable. In only 1 dog was subatmospheric pressure observed (−40 mm. of water). In most cases, the pressure fell steadily from whatever initial pressure was used to low levels in the range of 5 to 10 cm. above atmospheric pressure. In 2 instances, a column of saline 40 cm. high would not run into the duct, although slight pressure with a syringe served to force the fluid through. This was presumed to be due to spasm of the duct.

Attempts at measuring the pressure several days after cannulation, during which interval the heat-sealed end of the tube had been buried subcutaneously, usually showed the duct to be occluded. In 1 case cited above, the pressure, after 48 hours, was found to be 1 cm. of water.

These experiments indicated that flow of cerebrospinal fluid would occur from an obstructed ventricular system into the cannula. A series of 4 dogs, previously rendered hydrocephalic, was then successfully carried through the shunting operation. The results are shown in Table 2. Two additional animals were excluded from the series because of complicating operative infections.

TABLE 2

Animal No.	Re-explored	Ventriculostomy	Thoracic Duct	Autopsy
N-84	18 hrs. Functioning	O.K.	O.K.	Air embolism
N-128	8th day. No function	Tube out	Refluxed blood	Sacrificed
N-116	11th day. No function	Blocked	Refluxed blood	Sacrificed
N-89	35th day. No function	O.K.	Refluxed blood	Sacrificed

The occurrence of reflux (and clotting) of blood within the cannula in 4 of 6 cases, is at first sight the probable cause of failure of these shunts. However, in 2 of these, the ventriculostomy was not functioning. This suggests that the cannulae in these cases may have delivered fluid into the duct for only a short period, until the ventricular system was decompressed. In

2 instances in which an otherwise normal dog was subjected to cannulation of the thoracic duct, reflux of blood occurred, although in 1 of these the cannula has been demonstrated patent after 10 days. In the other, the clot was found at autopsy 10 days after cannulation. In a third similar experiment, the duct was patent 8 days after cannulation, as demonstrated at autopsy.

It is possible, as was suggested by the results of the ventriculo-thoracostomy series, that the great source of difficulty is the ventriculostomy. In this ventriculo-lymphangiostomy, there appears the further difficulty of ultimate incompetence of the valves at the entrance of the duct into the venous system.

SUMMARY AND CONCLUSIONS

As a further step in a general investigation of possible surgical treatment for hydrocephalus, polyethylene tubing of small caliber has been used to create shunts between the obstructed ventricular system and first, the pleural space, and second, the thoracic duct. These studies have led to the following conclusions:

(1) In the animals used in these experiments, the pressure within the ventricles during anesthesia was uniformly higher than that within the pleural space, or within the terminal segment of the ligated thoracic duct, as demonstrated by direct observation of movement of cerebrospinal fluid from the ventricles into the pleural space or into the thoracic duct.

(2) The pleural space is readily capable of absorbing isotonic saline solution instilled daily without losing its negative pressure.

(3) The thoracic duct does not conduct fluid other than lymph for more than a few days because of ultimate incompetence of the valves guarding its opening into the venous system, with subsequent reflux and clotting of blood.

(4) Until further improvements in technic and materials are made, ventriculo-thoracostomy and ventriculo-lymphangiostomy are not promising in dealing with human hydrocephalus.

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INTRACRANIAL ANEURYSMS

I. SOME CLINICAL OBSERVATIONS CONCERNING THEIR DEVELOPMENT*

ROBERT C. BASSETT, M.D.

Department of Surgery, University of Michigan Medical School and Hospital, Ann Arbor, Michigan

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THE fact that most aneurysms of the cerebral circulation are congenital in origin is general knowledge. Much evidence has been examined and inference and analogy have been drawn concerning the development of these lesions.[1,3,6,7,8,9,10,16] It seems to be generally agreed that in all likelihood defects within the muscular and elastic coats at angles of bifurcation of vessels eventually succumb to long-time persistent stress resulting in the formation of saccular dilatations.[6,7,8,9,10,16]

Less attention has been shown to the mechanism of development of the primitive cerebral vascular network offering through the complexity of its genesis a more simple and logical explanation for these anomalies.[1,17] Some writers, having noticed the presence of anomalous vessels, have by-passed the significance of these structures; others, however, have emphasized their importance.[1,5,6,9,11,12,14]

The following cases illustrate the point in question:

Case 1. F. M., 24-year-old white male. The aneurysm arose from the mid-portion of the left posterior inferior cerebellar artery and had blown out into the left cerebellar lobe. There were multiple anomalies of the surface and pial vessels (Fig. 1).

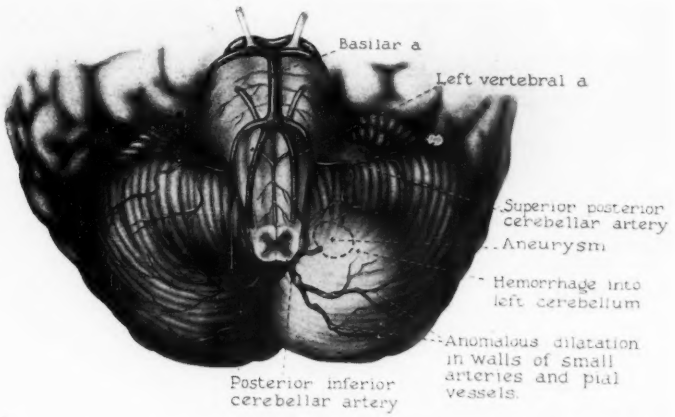
Basilar a
Left vertebral a
Superior posterior cerebellar artery
Aneurysm
Hemorrhage into left cerebellum
Anomalous dilatation in walls of small arteries and pial vessels
Posterior inferior cerebellar artery

FIG. 1. Case 1. Anomalous vascular stalk of aneurysm arising from left posterior inferior cerebellar artery. Multiple anomalous dilatations of pial vessels.

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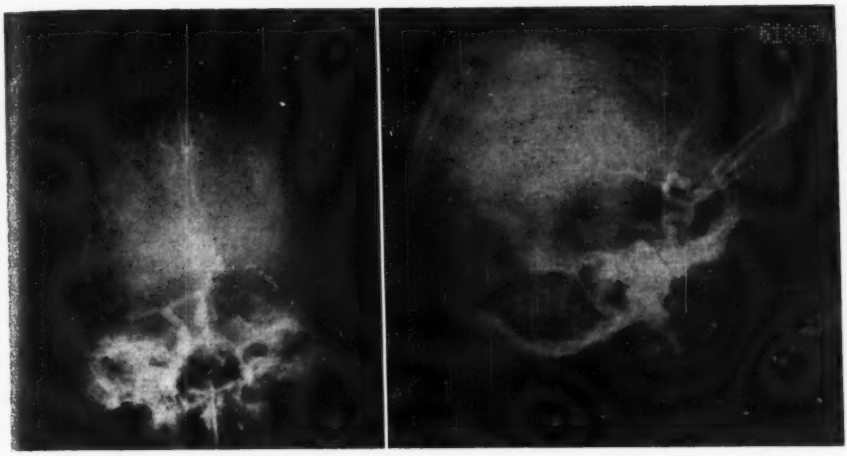


FIG. 2. Case 2. *Left*, AP view angiogram showing aneurysmal sac arising from mid-portion of anterior cerebral artery on the right between the anterior communicating artery and the carotid bifurcation. *Right*, Lateral view.

Case 2. M. Z., 40-year-old white female. The aneurysm was an elongated tortuous sac arising from the right anterior cerebral artery midway between the bifurcation and the anterior communicating artery, projecting forward and medially across the dorsum of the optic chiasm (Figs. 2 and 3).

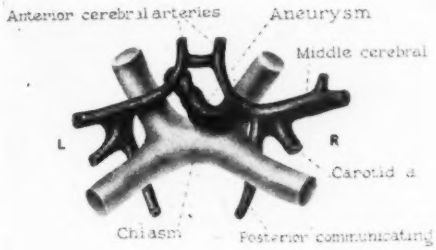
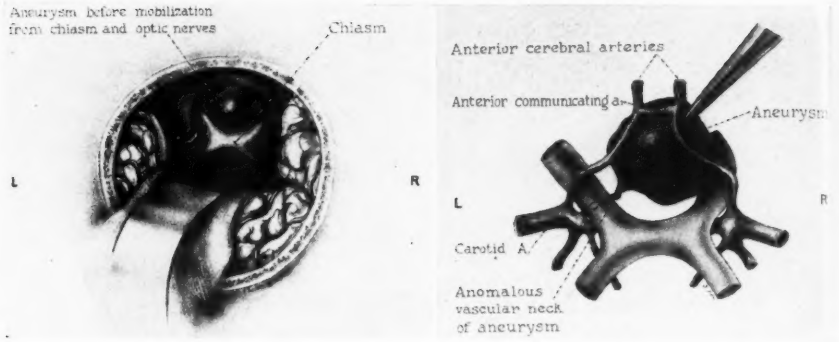


FIG. 3. Case 2. Diagrammatic reconstruction of aneurysm as seen at operation.

Case 3. F. B., 57-year-old white male. The aneurysm was situated within the sella turcica, elevating the chiasm and optic nerves to an angle of approximately 30°. The chiasm was almost completely bisected. The aneurysm represented a balloon-like termination of a tortuous anomalous vascular stalk arising from the medial wall of the left internal carotid artery beneath the left optic nerve (Figs. 4 and 5).

Case 4. G. R., 31-year-old white male. The aneurysm was a doubly lobulated structure arising from the mid-portion of the right anterior cerebral artery between the bifurcation and the anterior communicating artery. Hemorrhage had occurred from the smaller and more mesial of the two sacs to the left. Two anomalous vessels arose from the larger sac, one coursing superiorly with the right anterior cerebral artery and the other traversing the dorsum of the chiasm and dura of the floor of frontal fossa just to the right of the midline (Figs. 6, 7 and 8).

Case 5. C. S., 32-year-old white female. The aneurysm was situated below the



FIGS. 4 and 5. Case 3. Diagrammatic reconstructions. *Left,* Aneurysm as viewed at operation for suspected pituitary tumor. *Right,* Showing anomalous vascular source of aneurysm as seen at operation.

right optic nerve, elevating this structure. It extended medially, obscuring the diaphragma sellae, touched the left optic nerve and had its posterior limits just under the chiasm. It was almost completely thrombosed except for the portion of the sac near the tortuous neck, which arose from the mesial surface of the right internal carotid artery beneath the right optic nerve (Fig. 9). This situation was similar to that of Case 3 on the opposite side.

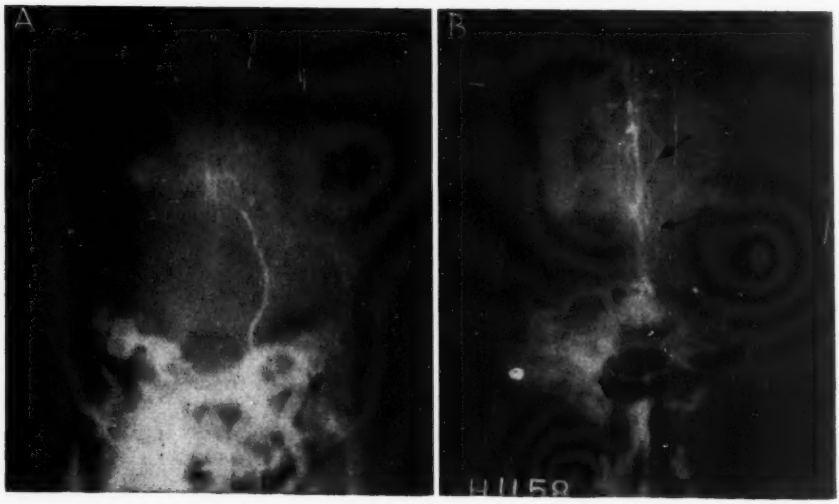
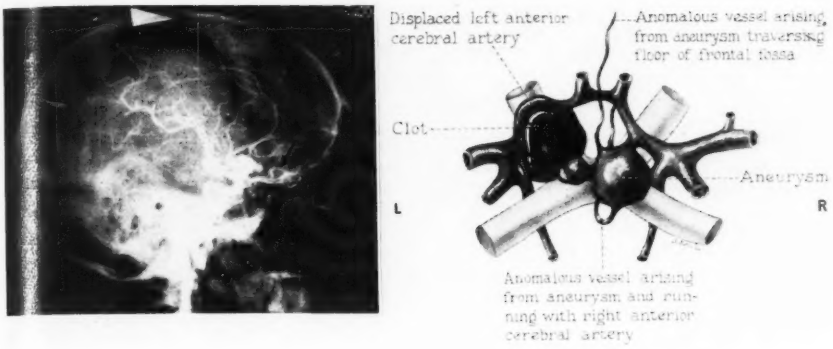


FIG. 6. Case 4. AP views. (A) Left-sided angiogram shows marked displacement to the left of the left anterior cerebral artery. (B) Right-sided angiogram shows bilobulated aneurysmal sac arising from right anterior cerebral artery and anomalous vessels which proved to arise from the sac.


Displaced left anterior cerebral artery
Anomalous vessel arising from aneurysm traversing floor of frontal fossa
Clot
Aneurysm
L
R
Anomalous vessel arising from aneurysm and running with right anterior cerebral artery

FIGS. 7 and 8. Case 4. *Left*, Lateral angiogram showing anomalous vessel arising from aneurysmal sac and traversing floor of frontal fossa. *Right*, Diagrammatic reconstruction of aneurysm and associated anomalous vessels as seen at operation.

DISCUSSION

As early as the 3 mm. stage in development of the foetus, vascular channels are well defined as such—containing primitive nucleated erythrocytes.[2,13,15] Definite morphological characteristics of artery and vein are undeveloped, a single endothelial layer forming the walls of the vessels.[15]

Vascularization occurs in response to metabolic demands from centers of proliferation.[4,15] Early the primordial endothelial channels give rise to innumerable buds forming a plexiform germinal circulatory bed. This gradually differentiates into efferent and afferent structures forming a fine capillary mantle.[15] Cleavage of vessel layers for skull, dura and brain next occurs, beginning in the basilar regions with multiple intricate anastomoses followed by adaptation to developmental alterations in form, size and rate of growth of the brain.[15]

Wyburn-Mason[17] has stressed this strategic relationship between proliferative vascular centers over and about the base of the developing brain with particular reference to the combined arterial and venous anomalies. This is especially true about the developing optic stalks[15] and is significant with respect to Cases 3 and 5 above.

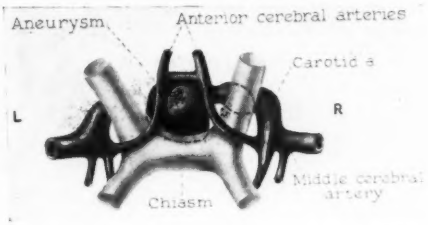


FIG. 9. Case 5. Diagrammatic reconstruction of aneurysm and associated vascular anomalous stalk as seen at operation.

Differential evolution to artery, vein, capillary and sinus types represents the final step in development. Not until mural structure is well developed are the arteries physiologically and anatomically

fixed as such, since there is much shifting about of arterial channels in young embryos.[11,15]

The development of the hyaloid artery to the primitive lens structure and its subsequent atrophy after capillary vascularization of the posterior lens capsule is a well known embryologic phenomenon suitably analogous to the question in point since this structure bears an intimate relationship to the proliferative vascular centers about the primitive optic pits, a site most commonly favored by aneurysms of the circle of Willis. This type of development, dissemination and atrophy of vascular channels where a specific need no longer exists is multiplied many times throughout the complex altering development of the brain and its vascular mantle (Cases 1, 2 and 4).

The carotid artery is the earliest branch of the primitive aortic arches ascending from the dorsum of the first arch to the midbrain, arborizing over the lateral aspects of this structure and anteriorly to the forebrain.[2,13,15] Its evolution is complete when atrophy of the connection between the dorsal aorta and the 3rd and 4th arches occurs.

Segmental dorsal aortic branches known as dorsal intersegmental arteries course between somites, the first pair atrophying early. The pair between the 2nd and 3rd somites remain longer, traveling with the hypoglossal nerve, and anastomose anteriorly with the internal carotids as paired primitive vertebral arteries, thus becoming a not too unlikely source for error in the development of the intradural aneurysms of the carotids below the circle of Willis. These primitive hypoglossal arteries soon atrophy and the vertebral arteries are formed by anastomosis of the 1st through the 7th cervical intersegmental arteries, which then fuse cephalad anterior to the brain stem to form the basilar artery.[2,11,13]

All of the above steps and fusion of the posterior portion of the circle of Willis through the posterior communicating arteries to the anterior portion through the internal carotid arteries have been completed by the 9 mm. stage (4 weeks).[4]

SUMMARY

Considering the embryological facts, congenital aneurysms of the cerebral circulation exist most likely as unresolved vestiges of a primitively normal circulatory system.

This primitive system has erred in its response to the evolutionary stimulus of resorption or modification for the pattern of normality as seen in the embryologically mature organism.

Five cases of congenital aneurysm of the cerebral circulation are offered as confirmatory evidence for this most likely mode of development.

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THE ROLE OF COMPLETE CEREBRAL ANGIOGRAPHY IN NEUROSURGERY*

RUPERT RANEY, M.D.,† AIDAN A. RANEY, M.D.,
AND J. M. SANCHEZ-PEREZ, M.D.
Los Angeles, California

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THE importance of establishing an exact topographic and pathologic diagnosis of an intracranial lesion cannot be overemphasized. Angiography, a valuable diagnostic procedure which at first met with opposition, is now generally acceptable and needs no further introduction. All intracranial lesions have some degree of influence on the vascular system; however, the effect is not necessarily restricted to the arteries alone; the arterioles, capillaries, and veins are often affected.

The average speed of blood flow from the common carotid artery in the neck to the internal jugular vein in the neck is 4½ seconds. This figure has been repeatedly computed by both chemical and roentgenographic methods.[4,5,26] Thus, a serial roentgenographic record of the movement of radiopaque material in the cerebral vessels during this interval of time offers an excellent opportunity for the study of vascular implications of various lesions.

Thorotrast and diodrast have been employed as contrast materials. Their advantages and disadvantages will not be discussed here, since they have been exhaustively explored by Northfield and Russell,[3,19] Egström and Lindgren,[6,7] Reeves and Stuck,[21,24] Lowman and Doff,[17] Martland,[18] and Gross.[10,11,12,13,14]

METHOD

The open operation or the direct intra-arterial puncture is used, depending on the indication. The nature of the lesion of course dictates the indication. The entire procedure is done on the X-ray table under sterile surgical conditions. An automatic seriograph is used which is adaptable to all standard X-ray equipment. This innovation developed by one of us (S.-P.) eliminates the confusion caused by apparatus requiring manual cassette changing reported previously by Moniz,[4,5] Sanchez-Perez,[22] and others. It allows reloading of films without interference to the operators. Controlled exposures are made .75 of a second apart by an automatic motor-driven cassette changer. Eight cc. of material are injected in a single burst through a 17 gauge needle.

Serial filming process is started just before the end of the injection.

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† 1136 West Sixth Street, Los Angeles 14, California.

Ordinary experience gives good timing. Six roentgenograms are taken within 4½ seconds at regulated time intervals. These films serially demonstrate the cerebral blood flow and constitute a reasonably complete angiographic record of the average patient. Film No. 1 usually shows the common carotid artery, the internal and external carotid artery, and the carotid siphon. Film No. 2 demonstrates the cerebral arterial system. Film No. 3 as a rule shows nothing but a diffuse opacity (this is the capillary phase), often important in neoplastic lesions. Film No. 4 is an intermediary stage and shows the ascending and descending veins. Film No. 5 demonstrates the venous elements emptying into the sinuses. Film No. 6 shows no dye in the cranial cavity in the average normal patient. In the event of pathologic conditions which prolong circulation time to an unusual degree, a second—third —ad finitum, series of films can be made. The second series would start 4–5 seconds after injection, etc., until the complete circulation time of the injected area is known. The instrument here reported is, therefore, not limited as far as time is concerned.

MATERIAL

Seven selected patients on whom angiographic studies were made have been chosen for report. They represent examples of lesions which differ considerably in their effects on cerebral circulation. The speed of blood flow in these patients has varied remarkably; it was either above or below normal depending on the nature of the lesion.

In the average patient with a vascular obstructive lesion, roentgenogram No. 6 will usually show the venous phase which indicates a slowing down of the blood flow. Conversely, in other patients with cerebrovascular short-circuit lesions, the opaque material leaves the cranial cavity more rapidly than in the normal case. It therefore becomes apparent that complete cerebral angiography can demonstrate not only topographic but pathologic data of increasing significance.

Arteriovenous communications, which have been described repeatedly, were recently reviewed by List[16] and illustrated by angiography. Such lesions are mainly of two types: (1) congenital vascular malformations and (2) arteriovenous aneurysms or fistulas of traumatic etiology. These arteriovenous lesions have been reported by Shenkin *et al.*[23] as factors that increase the rate of blood flow in the cerebral circulation. When the circulation time in the presence of such lesions is explored by chemical means, misleading conclusions are possible.

Certainly, the chemicals in the blood pass through the cranial cavity with greater speed because the way itself is shorter due to the short-circuited system. We believe, however, that under such circumstances the actual cerebral circulation time may be prolonged. It is true that a short-cut in the circulatory system allows the blood to leave the brain sooner, generally in 2½ seconds. The fact remains that the complete angiogram often shows no evidence whatever of opaque material entering the general cerebral circulation. On the contrary, the material usually remains confined to the short circuit.

Undoubtedly the reduced intra-arterial pressure above the lesion allows blood from other available sources to enter into the general circulation of the involved hemisphere provided the communication system is adequate. Thus the complete angiogram leads one to the conclusion that blood from a carotid artery involved in an arteriovenous lesion contributes little or nothing to the circulation of the affected hemisphere in many instances.

In a report by Govons and Grant[9] an angiographic illustration of a traumatic carotid cavernous sinus fistula showed opaque material not only in the fistula but also in the general cerebral circulation of the involved side (Case 11). Since there was no report on occlusion of the involved vessel, it could be inferred that the communicating arteries in the circle of Willis were either absent or too small to carry blood to the involved side. List and Hodges[16] state that such lesions usually cut out all dye to the general cerebral circulation.

Two patients with arteriovenous aneurysms, reported by Olivecrona in 1944 (cited by Sweet and Bennett,[25] p. 188) did not tolerate occlusion of the internal carotid artery but did tolerate ligation of the common carotid. The report was not available to us, so we do not know the size or location of these lesions. The communicating system in the circle of Willis, however, was undoubtedly inadequate, and therefore the external carotid artery was required as an accessory channel for satisfactory reflux collateral circulation.

The location of these short-circuit lesions varies considerably. When intimately related to the internal carotid artery or its siphon, a complete short circuit may well be anticipated. On the other hand, when the short circuit is in one of the cerebral vessels, particularly branches distal to the siphon, a complete short circuit probably will not obtain.

It is regrettable that good films were not obtained in some of the cases here presented. When the head support of the cassette changer was altered to facilitate reloading, the Lisholm grid was placed on top of the lucite cover. It was some time before we recognized that this change caused diffusion of the rays and lack of clarity in the films. These films, while satisfactory for clinical purposes, have not photographed clearly and have therefore been reproduced by drawings from the original films. The following cases serve to illustrate some of the short-circuit principles under discussion.

REPORT OF CASES

Case 1. Arteriovenous malformation distal to carotid siphon. Good collateral circulation through circle of Willis demonstrated by angiography. Circulation time delayed.

J. D., 0-660, a man of 35 years, was examined in May, 1941, because of syncopal episodes for 7 years. Subsequently attacks of motor aphasia developed (patient right-handed). They were of short duration. Later, Jacksonian convulsions occurred. Ultimately severe headache and coma followed. A right hemiplegia was present. A spinal tap gave bloody fluid. At operation an intracerebral hematoma (30 cc.) was found in the left inferior parietal area. In addition, there was a large arteriovenous malformation in the upper $\frac{3}{4}$ of the parietal lobe. He recovered, but a distressing

aphasia and right hemiparesis persisted. Further convulsive seizures continued even on anticonvulsant therapy.

The patient was re-examined on May 5, 1948. He had been having uncontrollable seizures with a postconvulsant maniacal state lasting several hours. A serial angiogram was done (5-11-48) which showed large arteriovenous channels, developing well above the origin of the middle cerebral artery. There was some evidence of blood entering the vessels contributing to the general circulation of the hemisphere (Fig. 1). The internal carotid artery on the side of the lesion was ligated. The patient made an uneventful recovery. There was some improvement of speech and con-

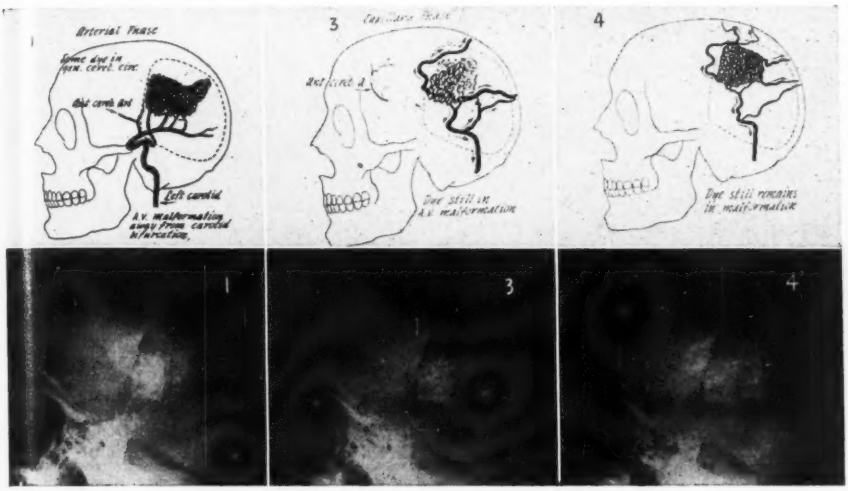

FIG. 1. Case 1. Arteriovenous malformation distal to carotid siphon. Some dye is in general cerebral circulation. A portion of dye is in anterior cerebral artery, as shown in Film 3.

vulsive seizures were controlled by anticonvulsant therapy; no further maniacal reactions have occurred.

Comment. The arteriovenous malformation in this case, while large, originated in the Sylvian group of vessels well away from the carotid siphon and an immediate by-pass of venous channels for the arterial blood was not present. Therefore it was possible for the carotid artery on the side of the lesion to contribute to the general circulation of the ipsolateral hemisphere through vessels not involved in the by-pass. This situation is especially true in smaller lesions (Elvidge,[8] Fig. 75, p. 137). Two angiograms by Hodes *et al.*, illustrating small frontal arteriovenous malformations, satisfactorily demonstrate this point (Cases 8 and 9).[15] A larger occipital lesion reported by Gross[14] further serves as an ample illustration. Thus, the improvement in symptoms after ligation of the internal carotid artery in this case indicates two probabilities: (1) adequate circulation to the involved hemisphere was received

from the circle of Willis and (2) the expansile qualities of the lesion were reduced.

Case 2. Congenital arteriovenous malformation at carotid siphon. Angiographic demonstration of complete by-pass of blood from ipsolateral carotid artery through lesion. No dye entered other vessels furnishing blood to involved hemisphere.

H. M., 0-356, a 33-year-old man, was examined, Sept. 19, 1939, because of spells of dizziness and black-outs since the age of 11. Weakness in the right arm and leg developed through the years. Numbness in the right hand occurred periodically,

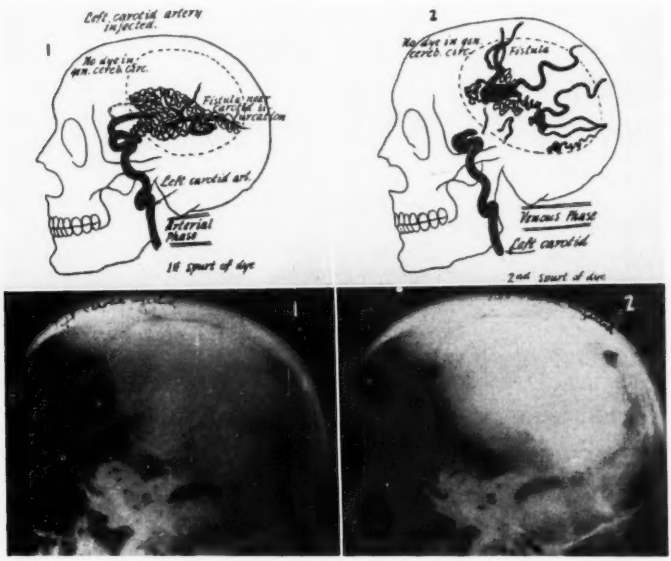

FIG. 2. Case 2. Lateral view illustrating short-circuit lesion. Dye is already entering longitudinal sinus in Film 2, taken approximately 1½ seconds after injection.

beginning first in the thumb. Jacksonian seizures appeared a year before examination and were associated with twitchings and paresthesias in the right face progressing down the right arm, leg and foot. There was an occasional momentary loss of consciousness at the termination of a seizure. Uncinate and gustatory fits commonly preceded the attacks. The blood pressure was 118 systolic and 74 diastolic. There was a mild hemiparesis on the right side (the patient was right-handed). Stereognostic sense was impaired in the right upper extremity.

Roentgenograms of the skull showed mottled calcifications low in the left parietal region. Operation, Oct. 4, 1939, disclosed a large arteriovenous malformation emanating from the Sylvian fissure. There were multiple ramifications and an excision was considered inadvisable. The patient had an uneventful recovery, and remained convulsion-free on therapy; petit mal attacks continued.

In June of 1947 the convulsions returned with greater frequency, even on therapy. Further there developed progressive loss of speech and increasing right hemiparesis. Roentgenograms showed no increase in the calcifications. An angiogram was done Mar. 25, 1948; it showed an enormous arteriovenous malformation originating near the carotid siphon. The dye was confined to the malformation and its passage time was shorter than normal (Fig. 2). There was no evidence of blood entering other vessels contributing to the circulation of the brain. The left internal carotid artery was ligated. Since the lesion crossed the midline an arteriogram was

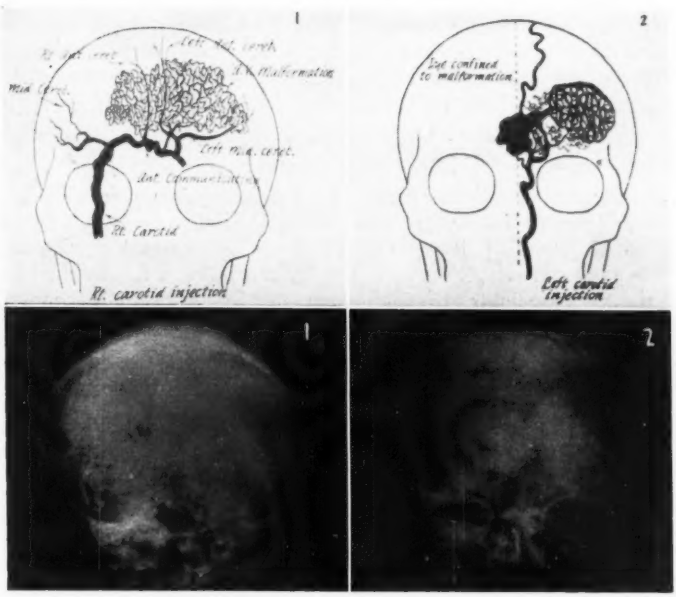

FIG. 3. Case 2. Contralateral carotid injection shows dye entering both hemispheres through collateral circulation.

done on the right side after the left carotid ligation. A large anterior communicating artery was seen and the opaque material entered into the circulation of both sides of the brain (Fig. 3). The patient again made an uneventful recovery; his speech returned and the hemiparesis cleared to a functional state. The stereognostic sense also returned to normal. While the time is not long there has been no return of convulsive seizures.

Comment. Serial angiograms of this patient showed no dye whatever entering the general cerebral circulation when injected on the side of the lesion. It therefore would appear extremely doubtful that blood from the ipsolateral carotid artery entered into the circulation of the involved hemisphere. The passage time of blood through the cranial cavity was shorter in

this case, undoubtedly because of the short circuit at the carotid siphon rather than to any material shortening of the actual cerebral circulation time. Ligation of the ipsolateral carotid artery caused no untoward effects. Actually, there was an improvement of symptoms after ligation. This indicates not only a reduction in the expansile qualities of the lesion, but also a collateral blood supply that undoubtedly served the involved hemisphere even before carotid ligation.

Finally, the later angiograms on the contralateral side, showing dye in both hemispheres, confirmed the impression gained by the first angiogram, namely, the presence of dye only in the lesion, indicating a complete short circuit and an adequate communicating source of blood through the circle of Willis to supply the involved hemisphere. The radical removal of three such short-circuit lesions by Pilcher[20] without serious neurologic deficit further indicates that these large vascular channels contribute little if any to the cerebral circulatory process.

Case 3. Traumatic carotid cavernous sinus fistula. Angiographic evidence suggesting a good collateral communicating system in circle of Willis.

A. S., 0-6809, a woman of 20 years, was thrown from a motorcycle and momentarily rendered unconscious, Mar. 21, 1948. She sustained a contusion to the left frontal and zygomatic areas and a bilateral compound fracture of the mandible. In addition, there was a fracture of the proximal third of the right humerus.

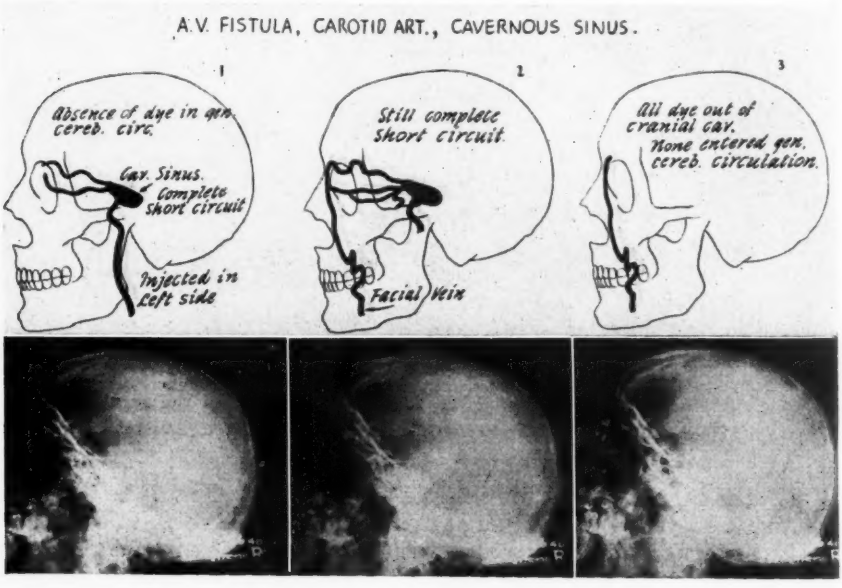

FIG. 4. Case 3. Note that all blood is out of cranial cavity in Film 3. The short time for passage of blood is due to short-cut lesions.

On the day after admission to the hospital she complained of diplopia and a swishing noise in the left ear. Examination showed a left 6th nerve palsy. One month later there was increased prominence of the left eye, with engorgement of the veins. Visual acuity was 20/20 in each eye. There was a persistent left 6th nerve palsy and a bruit was heard over the left fronto-orbital region, loudest over the orbit.

On May 20, 1948, serial angiography was done, which showed an arteriovenous fistula involving the carotid artery and cavernous sinus on the left side. No dye entered the general cerebral circulation; all dye left the cranial cavity in less than 3 seconds (Fig. 4).

On June 2, 1948, the left common carotid artery was ligated in continuity; this reduced the bruit and exophthalmos by 50 per cent. On June 9 the left internal carotid artery was ligated. The bruit and exophthalmos then disappeared. Furthermore, there was recovery from the 6th nerve palsy. Convalescence was not otherwise remarkable.

Comment. There are certain features in this case that may be of considerable interest. The fact that the patient had no cerebral symptoms during the months that the fistula was open indicates that cerebral circulation was maintained either through the ipsolateral carotid artery, or the contralateral carotid and basilar arteries through an adequate communication system in the circle of Willis. The angiograms, however, showed no dye entering into the general cerebral circulation on the side of the fistula. Therefore, it is reasonable to assume that the circulation of the involved hemisphere was maintained by blood from the circle of Willis and that the involved carotid artery was by-passed and contributed nothing to its circulation. Ligation of the internal carotid artery further substantiated the interpretation of the circulation in this case. Finally, angiographic findings such as shown here very likely indicate the presence of an adequate collateral circulation, and consequently serve as evidence of a situation essential for a successful trapping operation.

Case 4. Coma, a probable result of retarded circulation rather than a mild increase in intracranial pressure. Angiographic data of probable prognostic value demonstrate retarded circulation.

E. C., 0-6711, a woman of 68 years, was examined July 12, 1948, because of progressive loss of strength in the right hand for 2 weeks. Also there had been a complaint of increasing numbness in the right hand. A good history was not available because the patient was both aphasic and semistuporous. As far as could be determined from members of the family, none too familiar with the condition, there probably had been some change in personality for 2 or 3 months. Her past history was not especially remarkable. She had been quite active and had had no serious diseases or complaints known to available relatives.

On neurologic examination, the patient answered questions in either the affirmative or negative but did not engage in spontaneous conversation. She understood spoken language but rapidly fatigued and tended to go to sleep. The optic discs showed blurring of the margins and engorgement of the veins. There was a right central facial weakness and a right hemiparesis with some reduction in pain sensibility on the right side. Pathologic toe signs on the right were positive. Further reliable findings could not be obtained because of aphasia and stupor. Roentgenograms of

the chest and skull were normal. Lumbar puncture gave 250 mm. of water pressure. The fluid was clear and there were no other remarkable findings.

Since the character of the lesion was not certain, whether vascular or neoplastic, a serial angiogram was done July 15, 1948. The dye remained in the arterial vessels through the entire seriograms. There was slight forward displacement of the anterior cerebral arteries and poor filling of the Sylvian group (Fig. 5). In view of the increased intracranial pressure, an exploratory craniotomy was done July 16, 1948, which revealed acute swelling of the temporal lobe extending into the parietal lobe. There was venous thrombosis and extravasation of blood in areas through the involved temporal cortex. Similar changes were observed in the adjacent parietal cortex. A small artery in both the temporal and parietal regions was incised, which showed a sluggish stream of blood in each instance. Biopsies were taken and closure was done.

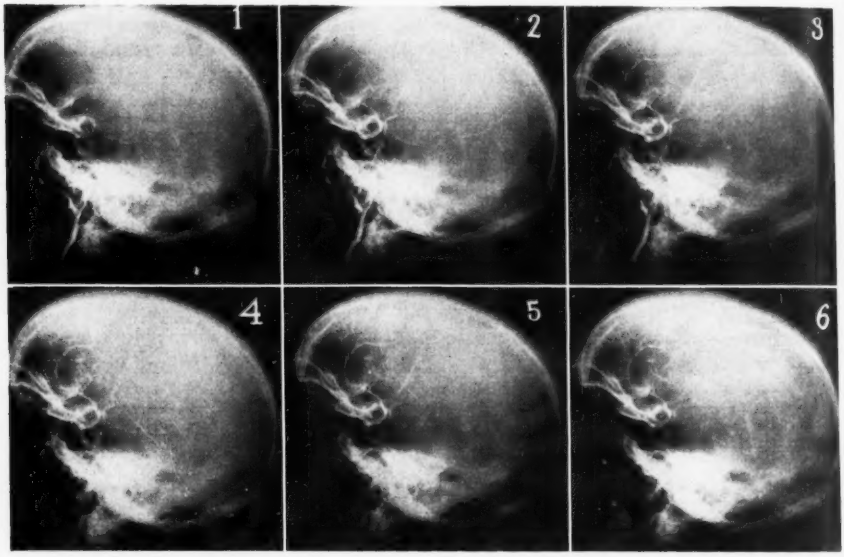

FIG. 5. Case 4. The complete seriogram shows retarded circulation. Blood is still in arterial system through the entire series of films. Lesion proved to be primarily a venous thrombosis and acute swelling in the temporoparietal region. This was the first seriogram taken with the finally improved instrument (35 per cent diodrast).

The patient's course was unchanged for a few days and then she expired suddenly. Autopsy revealed extensive venous thrombosis and acute swelling throughout the left parietotemporal area.

Comment. It is of interest here that the dye remained in the arterial channels throughout the normal circulation time of $4\frac{1}{2}$ seconds, there being little change in the location of the dye throughout the series of films. This indicated a generalized high-grade retardation of blood flow and consequently a

profound reduction in blood supply to the hemisphere. Thus it appears that coma was caused by a widespread reduction in blood supply instead of the moderate increase in intracranial pressure. Under such circumstances irreversible changes in the cortex are naturally prone to occur. Whether blood stasis occurs from pressure on venous outlets by space-occupying lesions or thrombosis, the end result is the same. Therefore, angiograms of this character showing a marked retardation of circulation suggest a less favorable prognosis regardless of the nature of the lesion or what is done. At operation the findings in this case suggested to us the picture of thromboangiitis obliterans so well described by Davis and Perret,[2] but were not verified by microscopic sections.

Case 5. Cerebral thrombosis with some features suggestive of a neoplasm. Angiographic evidence of lesion, retarded circulation, and probable invasion of anterior cerebral artery into ischemic area.

H. B., 0-6311, a man aged 60, was examined Mar. 6, 1948, because of increasing hemiparesis on the left side, and clouding of memory beginning 3 weeks previously.

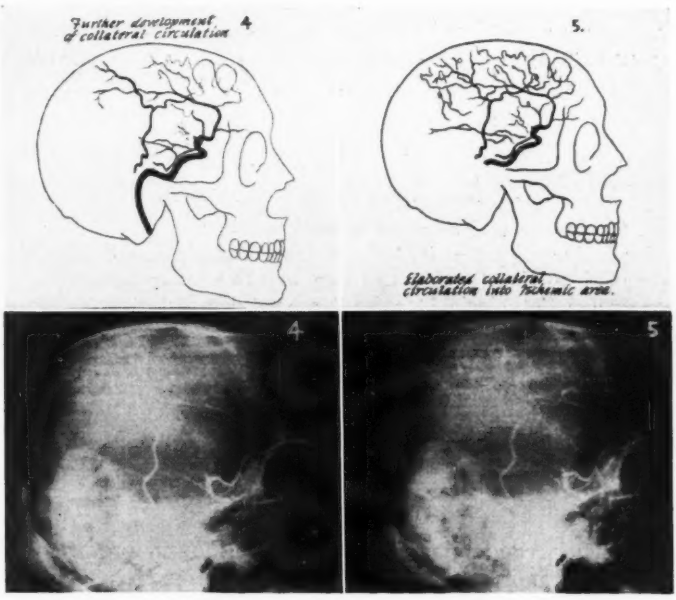

FIG. 6. Case 5. Thrombosis of middle cerebral artery. Films 4 and 5 suggest invasion of the anterior cerebral artery into the ischemic area. Temporal artery is visible. Dye in arterial system indicates general retardation of blood flow.

On examination, he was found to have a marked left hemiparesis and a cloudy mental state. On continued observation he showed some increasing weakness in the left side, but otherwise remained essentially the same. The patient was dull, apa-

thetic and at times stuporous. He finally showed a left hemiplegia with pathologic toe signs and a hemihypesthesia. The optic disks and the retinal vessels were tortuous. Visual field and other studies were unreliable because of the patient's condition. It was felt that he probably had a vascular lesion but the tendency toward progression of signs and symptoms suggested to us the possibility of a neoplasm. After observation for 11 weeks angiograms were done May 7, 1948, which showed a typical thrombosis of the middle cerebral artery on the right side. In addition the circulation time was slow. The dye was still in the arteries in the 4th, 5th and 6th films of the series (Fig. 6).

Comment. Improvement of symptoms following cerebral thrombosis, injury, etc., is well known. Further, following vascular insults of any kind collateral circulation tends to invade the ischemic area. Unfortunately for comparison no early seriograms were made in this case at the time of the thrombosis. The later films, however, strongly indicate that an elaboration of vessels into the ischemic area had taken place. The area originally supplied by the now thrombosed middle cerebral artery and its branches appears to have been invaded by the anterior cerebral artery (Fig. 6). Any increase in collateral blood supply that may have taken place probably occurred during the 11 weeks of illness. In addition to the area of ischemia, the general circulation time of the hemisphere was retarded, further contributing to his symptoms, particularly stupor.

Case 6. Angiographic demonstration of meningioma best seen in capillary phase and in addition evidence of retarded circulation to be reckoned with at operation.

A. McC., 0-6810, a man of 58 years, was examined on Feb. 2, 1947, because of periodic Jacksonian seizures of the left arm for 1½ years, often associated with pain in the arm. He further had progressive weakness in the left side for 15 months. Hypertension had been present for 2 years. On neurologic examination, there was a left hemiparesis involving the arm and face and, to a lesser extent, the leg. There was no sensory loss. Pathological toe signs were not demonstrated.

EEG done on Jan. 2, 1947, showed an abnormality in the right parietal region. Ventriculography, Jan. 27, 1947, showed a depression of the right lateral ventricle in the parietal area and a shift of the 3rd ventricle to the left. Jan. 31, 1947, serial angiograms were done, which outlined the tumor mass in the frontoparietal area (Fig. 7).

On Feb. 3, 1947, a first-stage operation was done, with a somewhat stormy course for the first 2 days. One week later the tumor was finally removed *in toto* (fibroblastic meningioma), with an uneventful convalescence and recovery from the hemiparesis.

Comment. The serial angiograms in this case showed a retardation of the circulation in the hemisphere as a result of obstruction to the venous outlets by the tumor. Further, the tumor was shown to its greatest advantage in what we believe was the capillary phase, as evidenced by the cloud of opaque material retained in the tumor. Of surgical interest, we believe that the delayed circulation time served as an indication of impending circulatory failure in the hemisphere. It is well known that after removal of benign tumors such as this the postoperative course is often stormy and all too fre-

quently fatal, regardless of the ease of the operation. It is further likely that impaired circulation in the hemisphere is the cause of these stormy courses. For this reason a multi-stage operation was done. On exposure, the tumor was decompressed but left *in situ*, thereby allowing the circulation of the hemisphere to adjust itself before proceeding further toward complete removal.

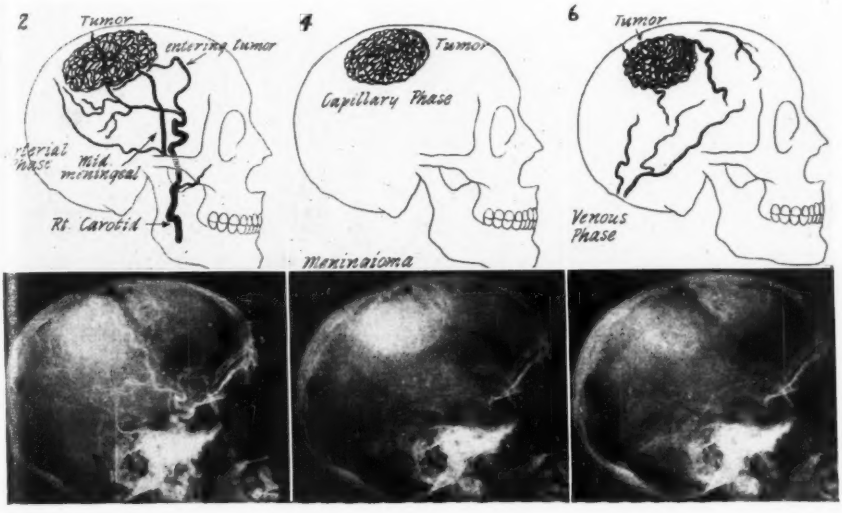

FIG. 7. Case 6. In Film 4 cloudy opacity is interpreted as capillary phase, which best demonstrates the tumor. Persisting dye in Film 6 indicates retarded circulation in hemisphere.

Case 7. Bilateral carotid aneurysms below siphon. Angiographic demonstration of cannulated portion. Successful surgical removal of lesion. Later death from rupture of sister aneurysm on other side. Notes on collateral circulation.

B. B. C., 9334, a man aged 57, was examined Sept. 30, 1947, because of increasing fatigability and emotional instability.

According to the history given by the wife, the patient had for several years shown a slight weakness in the left arm. In recent months it had increased to the point of disability. He had gained considerable weight. His memory became impaired and he showed marked emotional instability, crying or laughing in paradoxical fashion. He had sustained a head injury of undetermined severity at the age of 3. Otherwise the past history was not remarkable.

On neurologic examination, the patient was found euphoric and emotionally unstable. A left hemiparesis and bilateral Babinski signs were present. The abdominal reflexes were absent. The laboratory studies disclosed a negative blood Wassermann, and other routine studies were not remarkable.

An EEG, Oct. 7, 1947, revealed a focus of abnormality in the right parieto-temporal region. A pneumoencephalogram was done Oct. 17, 1947, which showed

no filling of the mid-portion of the right lateral ventricle. The visible ventricular pattern was not distorted. An angiogram revealed a rather amazing picture consistent with cannulated aneurysm (Fig. 8).

A craniotomy was done on Nov. 5, 1947, and a large aneurysm of the right internal carotid artery was trapped and removed without technical difficulty; it measured 7 cm. in its greatest diameter. The postoperative course was satisfactory for 36 hours. The patient responded and showed no change suggesting impaired circulation from sacrifice of the carotid artery. By the end of 36 hours, however,

FIG. 8. Case 7. Arteriogram illustrating cannulated portion of carotid aneurysm. [Film accidentally was photographed on wrong side, giving the appearance of a left-sided lesion. Actually the lesion was on the right side. Film was lost, making correction impossible.]

acute pressure symptoms suddenly developed, and he died within the hour. Autopsy revealed a symmetrically situated sister aneurysm on the opposite side, measuring 3 cm. in its greatest diameter, which had ruptured, causing the sudden demise (Fig. 9).

Comment. The relatively uncommon occurrence of multiple intracranial arterial aneurysms has been reported in numerous contributions. Dandy[1] in his monograph on arterial aneurysms describes these lesions in detail. Bilateral internal carotid aneurysms, often symmetrical, are not infrequently encountered. These lesions are of interest since their recognition is virtually dependent on bilateral angiography. The bilateral lesions in this comparatively unusual case, had we suspected them, could have been demonstrated by angiography. It is doubtful, however, that bilateral angiography is indi-

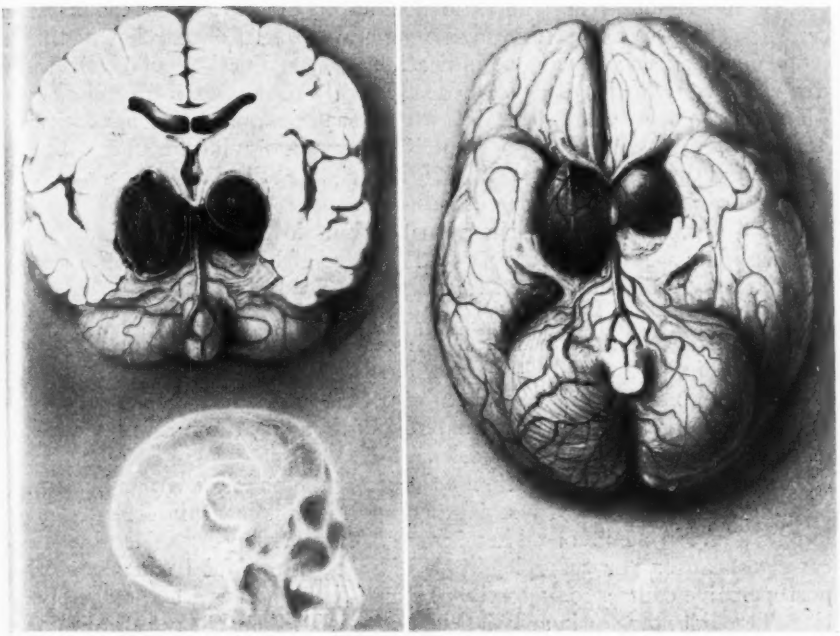

FIG. 9. Case 7. Artist's drawing showing approximate location of bilateral carotid aneurysms and phantom of angiogram.

cated in such a case. Even had we demonstrated the sister lesion, its removal probably could not have been accomplished without seriously interfering with essential collateral circulation. The circle of Willis, while intact, did not appear adequate to carry the necessary blood supply from the basilar artery had we trapped both of these lesions.

SUMMARY

From the foregoing data certain seemingly reasonable deductions have been drawn. In the case of congenital arteriovenous malformations distal to the carotid siphon, the ipsolateral carotid artery may contribute to the circulation of the involved hemisphere. Nevertheless, in most instances collateral circulation to the involved hemisphere would appear to be fairly well developed in these lesions.

In the case of large congenital arteriovenous communications at or near the carotid siphon, the ipsolateral carotid artery apparently contributes little or nothing to the circulation of the involved hemisphere, the circulation being maintained through adequate collateral communicating sources in the circle of Willis.

In the acquired or traumatic fistulas of the carotid-cavernous sinus variety, the presence of dye in the general cerebral circulation of the involved side is probably a good indication that collateral circulation through the circle of Willis is not developed sufficiently to tolerate a trapping operation. On the other hand, if no dye whatever appears in the general circulation of the involved hemisphere, it is pretty good evidence that collateral circulation through the circle of Willis is adequate and that a trapping operation can probably be done successfully.

In the presence of arteriovenous communications of all kinds, serial angiographic studies indicate a rapid passage of blood through the cranial cavity because of the short cut. Further, the blood in these short cuts contributes little, if any, to the circulation of the involved area. Exceptions are in those instances in which the lesion is well removed from the carotid siphon above the circle of Willis.

Although not proven, it would appear that in the case of cerebral thrombosis the uninvolved vessels are capable of invading the ischemic area to a considerable degree within 11 weeks.

Finally, a marked retardation of the cerebral blood flow, as can be demonstrated adequately only by serial angiography, indicates impending circulatory failure in the involved hemisphere. This may be of considerable surgical significance. For example, in the case of intracranial tumors, stormy postoperative courses may possibly be avoided by doing a two-stage operation. The first stage, primarily a decompression, favors the re-establishment of circulation. Then the tumor can be removed later with probably less danger.

In what we have interpreted as the capillary phase, the outline of certain tumors is more efficiently demonstrated. These lesions actually could be missed without a serial study, since angiograms done by the ordinary methods might very well not be taken at a time that the dye is suitably distributed.

When dealing with bilateral aneurysms of the internal carotid artery, bilateral angiography is usually essential to an accurate diagnosis. Its merit, however, would appear mainly academic since it is virtually impossible to sacrifice both internal carotid arteries in the average case without seriously jeopardizing cerebral circulation.

There are, of course, certain exceptional circumstances. For example, if bilateral lesions exist and only one is demonstrated, an attack on the demonstrated lesion might allow another more dangerous lesion to remain unsuspected and thus increase the probability of complications or a fatal issue.

Finally, a complete angiographic record of cerebral circulation taken in a controlled series offers an excellent opportunity for the study of pathologic conditions of the brain with altered circulation. Furthermore such information gained may be of utmost surgical importance when dealing with certain lesions.

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A NEW OPERATION FOR THE TREATMENT OF COMMUNICATING HYDROCEPHALUS

REPORT OF A CASE SECONDARY TO GENERALIZED MENINGITIS*

DONALD D. MATSON, M.D.†

Department of Neurosurgery, Duke University Hospital, Durham, North Carolina

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PATIENTS suffering from severe purulent meningitis may survive with the aid of modern antibiotic therapy only to face an equally disastrous complication, namely, obliteration of the subarachnoid fluid pathways by an adhesive process involving particularly the cisternae at the base of the brain and the subarachnoid spaces over the cerebral and cerebellar hemispheres. This chronic adhesive process may prevent access to, or virtually destroy, the normal absorptive surface provided by the vascular bed of the pia-arachnoid. The circulation of spinal fluid is therefore interrupted and progressive hydrocephalus ensues.

Hydrocephalus of this origin may or may not be of the so-called "communicating" variety, depending upon where interruption of the subarachnoid fluid pathways occurs. Several locations are involved commonly, and treatment varies accordingly:

(1) If one foramen of Monro is occluded, unilateral hydrocephalus may occur; this has been successfully treated by making an opening through the septum pellucidum.

(2) If the Sylvian aqueduct becomes blocked by scarring, but the subarachnoid cisternae remain open, subfrontal or subtemporal 3rd ventriculostomy may be performed, or even more satisfactorily, a shunting procedure of the type introduced by Torkildsen[9] which connects the lateral ventricle with the cisterna magna.

(3) If obstruction occurs in the posterior fossa alone with obliteration of the cisterna magna and occlusion of the caudal and lateral recesses of the 4th ventricle, then suboccipital craniectomy with lysis of these adhesions and removal of the thickened membrane which seals the 4th ventricle may be sufficient to re-establish normal fluid circulation.

(4) If, however, a more extensive adhesive process has taken place, causing obstruction of the basilar cisternae or the subarachnoid bed generally over the cerebral and cerebellar hemispheres, either alone or in addition to the locations already mentioned, then treatment becomes much more difficult. The operative procedures mentioned are of no avail since they fail to provide access to an absorptive surface. The problem becomes essentially the same as in congenital idiopathic communicating hydrocephalus where no

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† Now Associate Neurosurgeon, The Children's Hospital, and Junior Visiting Neurosurgeon, Peter Bent Brigham Hospital, Boston, Massachusetts.

obstruction can be demonstrated but there is apparently a simple failure of spinal fluid resorption.

Partial choroid plexectomy—for this can never be a complete operation—is the only procedure generally used in the treatment of communicating hydrocephalus. This involves at least two transcortical operations and the results are satisfactory probably only in patients where spinal fluid resorption is impaired to a minor degree.

Diversion of spinal fluid to a wide variety of locations has been carried out in the attempt to find an alternative absorptive surface. These locations include the subcutaneous tissues, the paranasal air sinuses, the pleura, the peritoneum, the thoracic duct and the blood stream itself, including both the intracranial venous sinuses and the veins of the scalp and neck. To date none of these has demonstrated any significant success either in hydrocephalic animals or in human patients.

If the search for a new absorptive area is abandoned, the possibility of shunting fluid into an excretory channel instead naturally comes to mind. With the exception of one or two disastrous attempts to lead fluid from the ventricles into the oro-pharynx and one attempt to suture a loop of bowel directly to the lumbar dura, the gastrointestinal tract has not been employed.[2]

The urinary tract has proved somewhat more attractive. Heile, in 1925,[4] first reported ureterodural anastomosis following unilateral nephrectomy. He operated on 4 patients; 1 died during the operation and the other 3 apparently were at least temporarily improved. Christopher in 1929[1] transplanted the ureter to the sac of a lumbar meningocele in 2 patients; 1 of these died at operation and the other 3 weeks later. Davidoff and Bancroft[3] reported 1 ureterodural anastomosis in 1932, the patient succumbing 1 week after surgery.

In 1934, Lehman[7] described 3 cases in which he had performed ureteroarachnoid anastomosis. The anastomosis no longer functioned at $7\frac{1}{2}$ weeks in 1, 1 died of meningitis at 17 days, and the third succumbed suddenly of unknown causes on the day of leaving the hospital. This same author collected reports of 13 individual cases from the Russian and German literature and estimated that 4 of them were in some measure successful. In 1942, Sachs[8] described 3 instances of uretero-arachnoid anastomosis performed following unsatisfactory results from choroid plexectomy; there was 1 death and the other 2 patients are reported improved. Torkildsen, in his recent monograph on ventriculocisternostomy,[10] dismisses uretero-arachnoid anastomosis as follows: "This ingenious operation is connected with a very high mortality and has never gained considerable practical importance because of its operative and post-operative complications."

This report presents an operative procedure which is an adaptation of the principle introduced by Heile, designed to shunt spinal fluid from the lumbar subarachnoid space to the urinary tract. Based upon considerable laboratory experience in the diversion of spinal fluid through various types

of channels[6] a method was adopted in this case which proved technically simple and which appeared to overcome many of the drawbacks of Heile's operation. The ease with which the procedure was carried out, the striking immediate recovery of this critically ill patient, and the uneventful postoperative course, with demonstration at $2\frac{1}{2}$ months that the excretion of spinal fluid by way of the bladder was still completely satisfactory, seemed to warrant preliminary presentation of this case in some detail.

CASE REPORT

An 8-year-old colored girl was injured in an automobile accident in May, 1947, at which time she suffered an extensive frontoparietal skull fracture. She was hos-

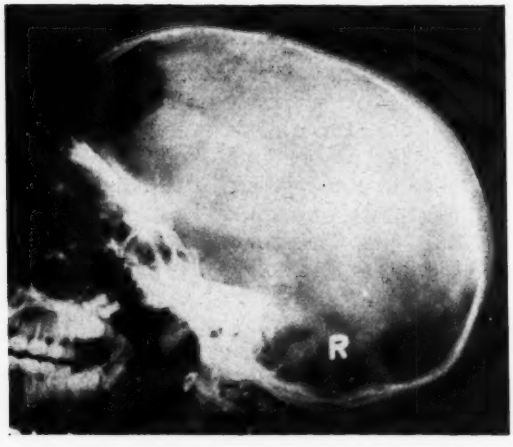


FIG. 1. Extent of craniectomy for removal of infected bone and repair of the dural defect.

pitalized for 3 months with gradual recovery, following which she was asymptomatic except for intermittent mild headache for 10 months—that is, until March, 1948. At that time she experienced the onset of severe headache, irritability, vomiting and fever; 36 hours later, she was admitted to a hospital in the midst of a generalized convulsion. She was subsequently transferred to the Neurosurgical Service of Duke University Hospital. On admission she was found to have staphylococcus meningitis, osteomyelitis of the frontal bone, and CSF rhinorrhea. A radical extirpation of the infected bone and frontal sinuses was carried out (Fig. 1) and the dural defect repaired with a pericranial graft.

After a stormy postoperative course, the infection cleared, but evidence of increasing intracranial pressure appeared and ventriculography demonstrated marked ventricular dilatation (Fig. 2). Posterior fossa exploration was therefore performed at which time the cisterna magna was found to be obliterated and the 4th ventricle obstructed by adhesive arachnoiditis (Fig. 3).

Following this operation the child did well, but after the 5th postoperative day the frontal decompression again bulged and the patient became drowsy and apa-

FIG. 2. Ventriculogram showing symmetrical dilatation of the entire ventricular system. There is no air in the basal cisternae or subarachnoid spaces over the cerebral hemispheres.

thetic. Removal of fluid by lumbar puncture always decreased the protrusion of the decompression. Phenolsulphonphthalein injected into the lateral ventricle appeared in the lumbar spinal fluid in good concentration in 5 minutes. However, only 7.5 per cent of the dye was excreted in 2 hours and less than 30 per cent in 12 hours.

An intravenous urogram demonstrated good excretion of diodrast by both kidneys. During this period it was necessary to perform lumbar removal of spinal fluid

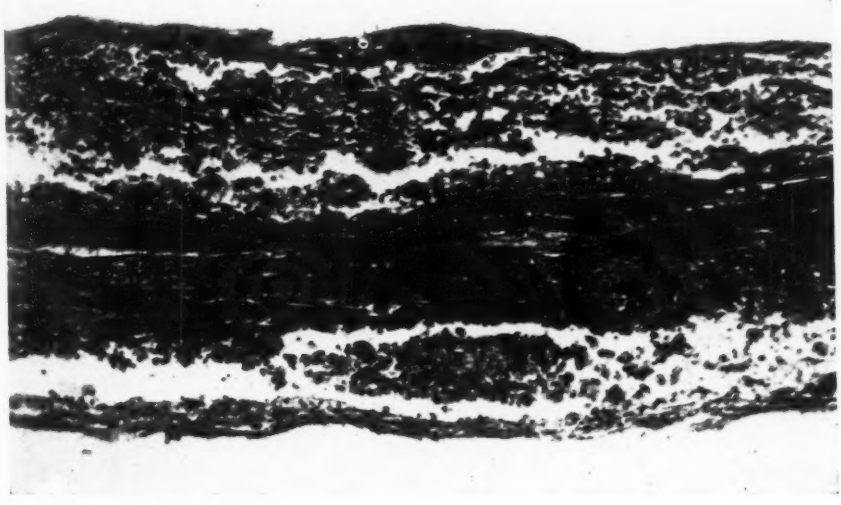


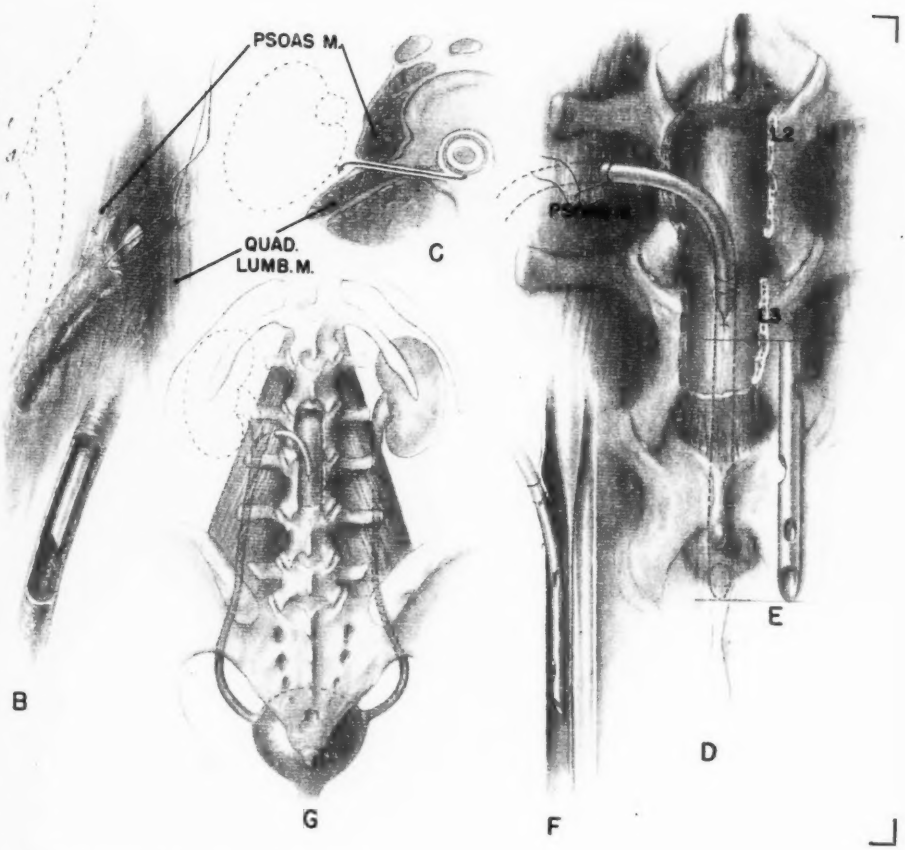
FIG. 3. Cross section of the membrane which sealed the 4th ventricle.

every 12 to 24 hours to relieve the patient of severe headache, semi-stupor, vomiting, and extreme bulging of the frontal decompression (Fig. 4). Immediately after each lumbar puncture she was much brighter and more comfortable but it became necessary to tap her at more and more frequent intervals. Since no absorptive surface within the central nervous system remained accessible, more radical measures were decided upon to relieve the increasing hydrocephalus.

FIG. 4. Extreme bulging of the intracranial contents through the bony decompression.

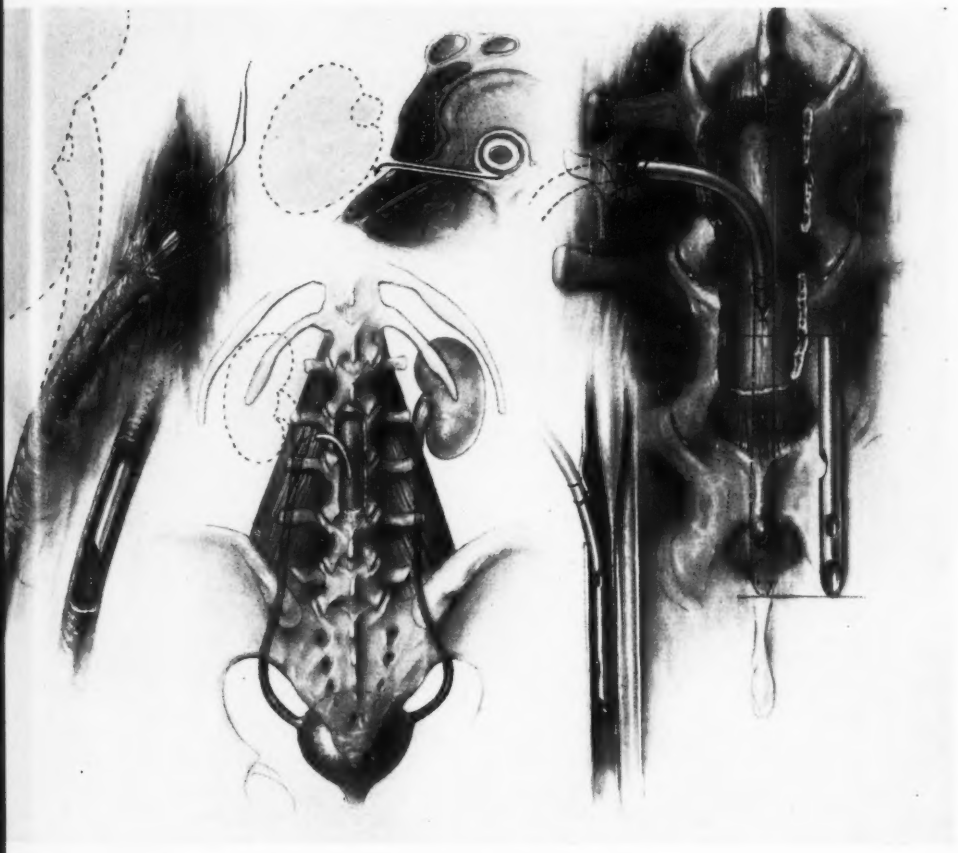
Operation

Choroid plexectomy was considered but the rapidity with which severe intracranial hypertension recurred following each lumbar spinal fluid drainage indicated a degree of obstruction which, it was felt, precluded any possibility of lasting benefit from partial plexectomy. On April 30, under ether anesthesia, the left kidney was removed through a flank incision by Dr. John Dees of the Urological Department. Leaving this incision open, the patient was rolled forward slightly and a laminectomy of L2 and L3 was carried out. The dura was opened in the midline. The arachnoid was punctured, and a polyethylene plastic tube,[5] about the size of a #8F catheter, was introduced and directed downward to lie among the cords of the cauda equina; approximately 6 cm. of the tube was placed intrathecally. The tube was fastened securely to the dura by silk sutures. It was brought upward and laterally toward the left in a gentle curve, tunnelled through the psoas muscle to the peri-nephric space and introduced into the ureter for about 6 cm. The margins of the upper end of the ureter were then sutured securely to the fascia of the psoas muscle. No sutures were placed around the ureter. Both operative wounds were closed without drainage (Fig. 5).



Detail of suture of the upper end of ureter to the fascia of the psoas muscle.
Detail of the plastic tube lying within the lumen of the ureter.
Cross section showing pathway of the tube from the spinal canal to the retroperitoneal space.
Laminectomy at L2 and L3, showing position of tube after insertion.
Detail of the end of the plastic tube inserted into the subarachnoid reservoir.
Relation of the tube to the dura and cords of the cauda equina.
General diagram of the completed operation.

5.



5. (A) Detail of suture of the upper end of ureter to the fascia of the psoas muscle.
(B) Detail of the plastic tube lying within the lumen of the ureter.
(C) Cross section showing pathway of the tube from the spinal canal to the retroperitoneal space.
(D) Laminectomy at L2 and L3, showing position of tube after insertion.
(E) Detail of the end of the plastic tube inserted into the subarachnoid reservoir.
(F) Relation of the tube to the dura and cords of the cauda equina.
(G) General diagram of the completed operation.

The *postoperative course* was afebrile. The patient was incontinent for about 36 hours and after that had complete urinary control. The decompression remained depressed at all times (Fig. 6). It was never necessary to tap the patient after this operation because of increased pressure. One lumbar puncture was carried out on the 5th postoperative day, at which time the initial pressure was 90 mm. of water.

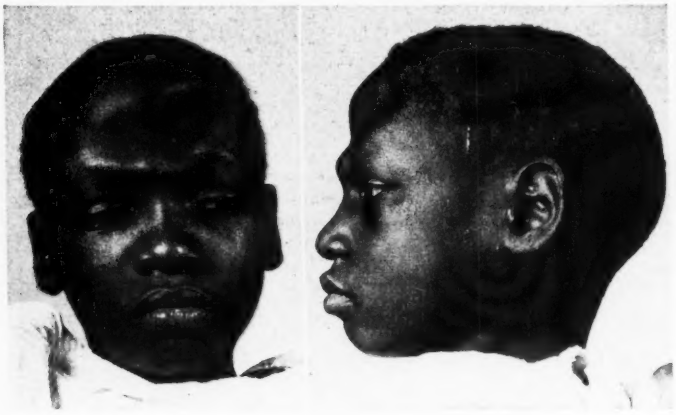


FIG. 6. Appearance of the decompression which persisted at all times after shunting spinal fluid from the lumbar subarachnoid reservoir to the left ureter.

Recovery was remarkably rapid. All wounds healed per primam and the patient, for the first time in over 8 weeks, took a real interest in her surroundings, laughed, ate well and wanted to get up. Her voided urine showed rbc. for 10 days and none thereafter. It continually showed 5–20 wbc./high power field in a centrifuged specimen. It showed no sugar, a 1+ albumin, and a specific gravity varying between 1.002 and 1.014.

On May 18, the coronal scalp flap was re-elevated and a preformed tantalum plate inserted to close the defect in the frontal bones (Figs. 7 and 8). The postoperative course was uneventful. She was up and about the ward by the 5th day and was discharged home on her 76th hospital day, 30 days after anastomosis of the lumbar subarachnoid space to the ureter and 10 days after insertion of the tantalum plate. At this time she was alert, afebrile and symptom-free.

The patient was readmitted to the hospital for evaluation 2½ months after the shunting operation. She had gained several pounds in weight, was happy and active. She had complete control of her urine, voided 4 to 6 times during each day and not at all during the night. Her neurological examination was normal. Her urine showed a specific gravity of 1.006, no albumin or sugar, and 6–10 wbc./high power field in a voided specimen. Lumbar puncture revealed an initial pressure of 50 mm. of water in the lateral position. Ventricular puncture was then carried out and the pressure in the dependent ventricle in the lateral position was also 50 mm. of water; 1 cc. of neutral phenolsulphonphthalein was injected into the ventricle. This dye appeared in the bladder urine in between 15 and 20 minutes; 75 per cent of the dye was excreted within 2 hours and approximately 95 per cent in 12 hours. Six months after operation, the patient is still symptom-free and leading a normal active life.

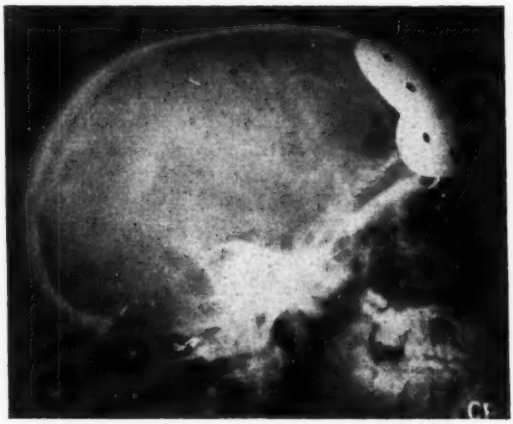


FIG. 7. Preformed tantalum plate inserted to close the bony defect.

DISCUSSION

This procedure was designed to provide several features felt to be desirable: first of all, ureteral peristalsis should remain unimpaired since the nerve and blood supply of the ureter was left intact; secondly, the normal valvular action of the uretero-vesical junction should also remain unimpaired since this region was not even approached; thirdly, the possibility of retrograde infection travelling from the bladder to the subarachnoid space by way of the lymphatics in the wall of the ureter should be obviated by the interpolation of a loop of polyethylene tubing at least 6 cm. in length between the ureter and the arachnoid. In the fourth place, the possibility of arachnoid adhesions occluding the junction of an artificial opening made into the subarachnoid space—which was frequently the fate of direct uretero-arachnoid anastomosis—was counteracted by placing a full 6 cm. of a fairly large size plastic tube containing several openings well into the lumbosacral subarachnoid reservoir and well away from any points of operative interference with the arachnoid. In the fifth place, a large enough loop of polyethylene tube was used, and a sufficient length of the tube inserted both into the ureter and into the subarachnoid space to allow any type of motion or position-

FIG. 8. Appearance of patient 8 days after tantalum cranioplasty.

ing of the patient without danger of the tube being dislodged or put under tension.

There was no evidence that this patient's electrolyte, carbohydrate, or protein metabolism was significantly disturbed by the continual excretion of large amounts of spinal fluid. This amount was not determined quantitatively since it was not felt justified to run the risk of ureteral catheterization. The consistently low specific gravity of the voided urine, the rapid excretion of the dye injected into the ventricular system, and more important perhaps, the clinical course of the patient seemed ample proof of the continued flow of spinal fluid through the artificial shunt.

There was no evidence that the persistent reduction in the pressure in the subarachnoid fluid spaces, measured at 50–90 mm. of water on two occasions, had any deleterious effect. Because of the rather alarming manner in which the frontal decompression sank in when the patient assumed an erect position, especially after she had just voided, it was felt that repair of the cranial defect at the earliest possible date might be desirable to lessen, perhaps, the possibility of subdural or subarachnoid hemorrhage. Therefore, not without some misgivings, tantalum cranioplasty was carried out only 60 days after the original removal of the osteomyelitic bone, exenteration of the frontal sinuses and grafting of the large dural defect. A satisfactory result was achieved and to date there has been no evidence of unusual reaction to the plate.

The history of the treatment of hydrocephalus is replete with reports of individual cases in which some new surgical procedure resulted in temporary improvement, only to have failure ensue later. Therefore, this should be viewed strictly as a preliminary observation. The subsequent course of this patient will be followed with considerable interest and care before any final conclusion is reached.

The following opinions, however, seem tentatively justified on the basis of the case presented:

(1) Combined nephrectomy and laminectomy for this purpose is not technically difficult or unduly shocking to the patient when the proper supportive measures are anticipated. In the case reported it was apparently a life-saving procedure.

(2) There was no undesirable disturbance in electrolyte, protein or carbohydrate metabolism as a result of the continual excretion of spinal fluid.

(3) No urinary dysfunction such as incontinence, frequency, nocturia, or painful urination has resulted from the mixture of spinal fluid and urine in the bladder.

(4) No abnormal neurological symptoms or signs have resulted to date from the presence of the plastic tube in the subarachnoid space amid the cords of the cauda equina.

(5) There is no evidence that any reflux of urine from the bladder to the subarachnoid space through this system has taken place in a patient who has been very active and led an apparently normal existence.

(6) If this patient's course continues as satisfactorily as it has proceeded to date, without evidence of increased intracranial pressure, foreign body reaction or meningeal infection, it is felt that this procedure should be explored further and applied to selected patients with communicating hydrocephalus of all types, especially those who have persistent increased pressure following choroid plexectomy.

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ADDENDUM

Since the preparation of this report, this operation has been performed without difficulty on 3 hydrocephalic infants, 3 to 5 months of age, at The Children's Hospital, Boston. Preliminary observations indicate that in infants 2–3 grams of salt daily should be added to the diet to prevent excessive chloride loss.

CASE REPORTS AND TECHNICAL NOTES

SPINAL EXTRADURAL CYST

HENRY G. DECKER, M.D.,* AND KENNETH E. LIVINGSTON, M.D.†
Department of Neurosurgery, The Lahey Clinic, Boston, Massachusetts

(Received for publication May 13, 1948)

The purpose of this report is to record another case of spinal extradural cyst.

The origin of these cysts is not altogether clear but in general they can be considered as derived from the spinal meninges and may be classified as: (1) parasitic cysts, which have been discussed in some detail by Krauss;[9] (2) teratomatous cysts, which have been described by Adams and Wegner,[1] and the so-called congenital type of cyst of the spinal meninges described by Cloward,[3] Mayfield and Grantham,[10] Elsberg, Dyke and Brewer,[5] and others. In addition, there are the traumatic extradural cysts, usually following operative procedures.[7,13]

A number of authors[4,5,8,11,12,14,15] have commented upon the fact that spinal extradural cysts are not a common cause of spinal cord or cauda equina compression. Adelstein,[2] however, has reported 17 cases and Turner[15] has reviewed 10 additional cases in which spinal cord compression did occur. Subsequent to Turner's report, Hyndman and Gerber[7] reported 3 cases and Swanson and Fincher[13] reported 3 additional cases of spinal extradural cyst.

REPORT OF CASE

A 31-year-old woman, a presser, was first seen at the Lahey Clinic on Aug. 28, 1947, complaining of paresthesias in the right knee and thigh. About 6 years before entry she began to notice some decrease in sensation in the region of the right knee. More recently she had suffered attacks of pain in this area and in the right hip, brought on particularly by kneeling or bending and frequently lasting 1 or 2 days.

Examination. The patient was well developed and nourished. The lower back was held stiffly and there was voluntary limitation of movement. Actual radiation of pain on movement was not elicited. There was an area of complete anesthesia over the anterior right thigh in the 2nd and 3rd lumbar dermatomes. There was slight quadriceps atrophy and weakness of extension at the knee. The right knee jerk could not be elicited. Blood Hinton and Wassermann tests were negative. Roentgenograms of the spine showed some reaction about the sacro-iliac joint, more marked on the left. The articular facets at the lumbosacral joint were asymmetrical, with slight scoliosis convex to the left.

On Oct. 27, 1947, pantopaque myelography was carried out. Dynamics were normal; total protein of fluid, 70 mg./100 cc.; Davies-Hinton and Wassermann reactions negative. Fluoroscopy showed the pantopaque flowing freely from the cul-de-sac to the mid dorsal levels. At the level of the 3rd lumbar vertebra on the right there was an extravasation of pantopaque into an area slightly posterior to the main column. The amount of extravasation increased each time the pantopaque was allowed to pass this point and finally measured 1.5 cm. in width and 3 cm. in length (Fig. 1). It was concluded that this lesion was a communicating cyst. Following fluoroscopy all of the pantopaque was removed except that retained in the cyst since this could not be brought down into the cul-de-sac. At the suggestion of the

* 503 Equitable Bld'g,, Des Moines 9, Iowa.
† 806 S. W. Broadway, Portland 5, Oregon.

radiologist, the patient was allowed to be ambulatory for the next 24 hours in order to observe whether the cyst might empty itself into the general subarachnoid space. On the following day the roentgenogram showed partial emptying of the cyst with accumulation of the pantopaque in the cul-de-sac (Fig. 2).

FIG. 1 (*left*). Pantopaque flowing into the extradural cyst during fluoroscopy.

FIG. 2 (*right*). Residual pantopaque after 24 hours. A portion of the pantopaque has left the extradural cyst to reaccumulate in the cul-de-sac.

Operation. On Nov. 1, 1947, exploration of the lumbar canal revealed beneath the lamina of the 3rd lumbar vertebra on the right a distended extradural cyst, about the size of a man's thumb, adherent to the dura. The cyst was opened during the process of dissection and was found to contain spinal fluid and material characteristic of pantopaque. It contained probably the 3rd right lumbar root which was adherent to the cyst wall. A silver clip was applied to the narrow neck of the sac and the cyst was removed, following which gelfoam was placed in its bed.

The pathologic microscopic report was cyst with fibrous degenerated walls.

Postoperative Course. The anesthetic area of the right thigh remained unchanged and the right knee jerk again could not be elicited. The wound healed well and the patient was relieved of pain. She was last seen 4 months following operation, at which time she stated that she was entirely free of pain and had no complaints. Sensory changes remained the same and the right knee jerk could not be elicited.

COMMENT

There was no evidence that trauma played any role in this case. The rather long history of sensory changes in the region of the right knee suggests that the cyst was congenital in origin, similar to cases reported by Robertson and Graham,[11] Elsberg, Dyke and Brewer,[5] and Cloward and Bucy.[4] Partial communication of the cyst with the subarachnoid space was present, similar to that noted in the case presented by Good, Adson and Abbott.[6]

In the presence of segmental sensory, motor and reflex changes, such as were present in this case, together with radiographic findings suggestive of an extradural cyst, the diagnosis of such a lesion should be considered as probable. In the patient reported here we believe that the appearance of pain symptoms coincided with distention of the cyst and that periodic subsidence of symptoms occurred when the cyst had been decompressed by drainage into the subarachnoid space.

CONCLUSION

We have presented another case of spinal extradural cyst. In this instance partial communication with the subarachnoid space could be clearly demonstrated by roentgenologic examination.

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SPINAL EXTRADURAL CYST

CASE REPORT AND TABULATION OF PREVIOUSLY REPORTED CASES

CAPTAIN COURTLAND H. DAVIS, JR., M.C., A.U.S.
Army Medical Center, Washington, D. C.

(Received for publication June 10, 1948)

Since the impetus of the reports of Elsberg, Dyke, and Brewer,[2] and Lehman,[7] a total of nearly 30 cases of non-traumatic, non-parasitic spinal extradural cyst have been recorded; the purpose of this communication is to present an additional case and to bring up to date the tabulated summary of Adelstein.[1]

CASE REPORT

An 18-year-old colored male, Pvt., U.S.A., was admitted to Walter Reed General Hospital on 27 Mar. 1947, after evacuation through channels from an overseas station, with chief complaint of progressive numbness, weakness and stiffness of both lower extremities of 9 months' duration. The patient stated that he also had mild difficulty in keeping his balance with his eyes closed, and he had gross ankle clonus ("when I step on my feet they jump"). At no time was there any complaint of pain, nor was there any bowel or bladder dysfunction. Family and past history were non-contributory.

Examination. He was a short, obese colored male, in no acute distress, walking with a moderately spastic gait. Romberg was positive. Examination of the spine revealed no limitation of motion, no paravertebral muscle spasm, and no tenderness to pressure or percussion; there was a moderate lumbar lordosis but no kyphosis. Straight leg raising was not limited; there was no gross atrophy of either lower extremity. The knee and ankle jerks were present and equal but they could not be interpreted as being hyperactive; abdominal reflexes were absent bilaterally; cremasteric reflex was present on the left but absent on the right. There was sustained ankle clonus on the right, unsustained on the left; Babinski sign positive bilaterally. Heel-to-knee test showed moderate incoordination bilaterally; however, position and vibratory senses appeared intact. Touch and pain sensations were present in both lower extremities and in the lower trunk, but there was a slight increase in these sensations at and above the costal margin bilaterally. Examination of the cranial nerves and upper extremities revealed no abnormality.

Routine blood and urine studies were negative. Roentgenograms of the dorsal spine revealed definite widening of the interpedicular space of the 7th dorsal vertebra with erosion and flattening of the pedicles, being more marked on the left.

On 10 April lumbar puncture was performed with patient lying on his side; initial pressure was 183 mm. H_2O; bilateral jugular compression caused rapid rise to 240 mm., but there was no fall following release of compression. Myelography was immediately performed, revealing a complete block of regular outline at the inferior border of D7. These procedures caused no undue distress nor change in the patient's condition. Spinal fluid protein was 192 mg. per cent.

Operation. Laminectomy of D7 and parts of D6 and D8. There was no extradural fat, and the exposed area was completely filled with a smooth, well demarcated, blue-gray, cystic mass, having the same gross structural appearance as normal dura. This was incised in the posterior midline, allowing escape of normal cerebrospinal fluid in which could be noted traces of the oily contrast medium that had been used for myelography. In the caudad portion could be seen a 2 mm. free communication with the spinal subarachnoid space. (This opening was at about 11:00 o'clock on the spinal dura and not near the emergence of a root.) The cyst wall was then easily dissected away from the dura, and the pedicle was tied off and cut; the size of the cyst was about $1 \times 1 \times 4$ cm. It was then noted that the dura, which had been markedly compressed, was hard and fibrosed. It was therefore opened in the midline for the entire

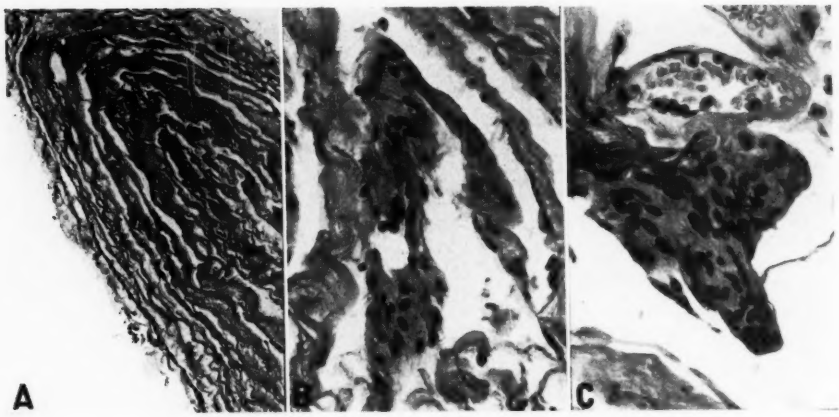

FIG. 1. Photomicrographs of spinal extradural cyst (hematoxylin and eosin stain). A, Collagenous cyst wall (×94). B, Delicate lining membrane (×266). C, Area of cellular proliferation (×266).

length of the exposure, with bulging out of the spinal cord. At the end of this procedure there was free communication with the subarachnoid spaces above and below. The dural edges could not be approximated; the wound was carefully closed in layers with silk.

Pathological Report. "*Gross:* Specimen consists of a delicate white cystic sac measuring 2 cm. in diameter and less than 1 mm. in thickness. The inner surface is smooth, white and glistening; the outer surface presents delicate blood vessels and tags of connective tissue. *Microscopic:* Cyst wall is composed of acellular dense collagenous connective tissue. There is a delicate lining membrane resembling endothelium which in several areas gives rise to a proliferation of epithelial-like cells morphologically similar to the meningothelial cells of the Pacchionian granulations." (See Fig. 1.)

TABLE 1

(*Adapted from Adelstein*)[1]

Case No.	Date Reported	Author	Age	Sex & Color	Duration of Symptoms	Location of Cyst	Dilation of Spinal Canal	Results & Comments
1	1898	Schlesinger	?	?	?	Midthoracic	?	Died of multiple sclerosis; postmortem findings.
2	1907	Krauss	46	M	13½ mos.	D3–D6	?	Progress arrested; patient invalid.
3	1932	Mixter	26	M	13 yrs.	D3–D7	?	No improvement
4	1934	Elsberg *et al.*[2] Case 1	12	M	3½ yrs.	D5–12	Present (x-ray)	Marked improvement
5	1934	Elsberg *et al.*[2] Case 2	15	M	3 mos.	D6–9	Present (x-ray)	Cure
6	1934	Elsberg *et al.*[2] Case 3	15	M	9 mos.	D4–9	Present (x-ray)	Slight Improvement
7	1934	Elsberg *et al.*[2] Case 4	16	F	2 yrs.	Midthoracic	?	Almost complete cure

TABLE 1—(*continued*)

Case No.	Date Reported	Author	Age	Sex & Color	Duration of Symptoms	Location of Cyst	Dilation of Spinal Canal	Results & Comments
8	1935	Lehman[7] Case 1	12	M, C	3½ mos.	D6–8	Present (op.)	Cure, kyphosis
9	1935	Lehman[7] Case 2	17	M, C	9 mos.	D6–10	?	Cure, kyphosis
10	1936	Cloward	43	M, W	2 yrs.	D10–L5	Present (x-ray)	Marked improvement
11	1936	Peet & Kahn	12	F, W	2 mos.	D9–L4	Present (op.)	Cure
12	1937	Kelly	15	M	5 mos.	D6–8	Present (x-ray)	Cure
13	1937	Cloward & Bucy	20	M W	6 yrs.	D7–9	Present (x-ray)	Marked improvement
14	1937 (1942)	Mayfield & Grantham[8]	16	M, W	3 yrs.	D6–10	None	Cure, kyphosis
15	1937	Teachenor						
16	1939	Robertson & Graham	14	M, C	6 mos.	D7–9	Present (x-ray)	Cure
17	1941	Adelstein[1]	15	F, W	5 mos.	D5–7	Present (x-ray)	Cure, kyphosis
18	1939	Turnbull[12]	18	F	4 yrs.	D6–10	Present (x-ray)	Cure
19	1942	Mayfield & Grantham[8]	26	M, C	12 yrs.	D6–7	Present (x-ray)	Cure
20	1942	Meredith[9]				Cervical		
21	1942	Gros[4]	14	M	4 wks.	D4–9	Present (x-ray)	Cure
22	1942	Hartmann[5]	14	M	1½ yrs.	D10	Present (x-ray)	Cure
23	1944	Good *et al.*[3]	42	M	10 yrs.	D8–L1	Present (x-ray)	Gradual improvement
24	1945	Shenkin *et al.*[10]	51	F	3 mos.	D7–9	Present (x-ray)	Cure
25	1945	Shenkin *et al.*[10]	13	F	2 yrs.	D6–8	Present (x-ray)	Cure
26	1946	*Hyndman & Gerber[6]	13	M	8 mos.	D4–10	Present (x-ray)	Marked improvement
27	1947	Swanson & Fincher[11]	20 mos.	F	Acute	L2–5		Marked improvement
28	1947	†Swanson & Fincher[11]	43	F	?	T12–L2		Cure
29	1948	Davis	18	M, C	9 mos.	D7–8	Present (x-ray)	Cure

* Also reported 2 cases acquired cyst.
† Also reported 3 cases acquired cyst.

Postoperatively the patient was fitted with a back brace as prophylaxis against kyphosis. His course was entirely uneventful with gradual diminution of previous symptoms. On discharge 3 months later he complained only of slight fatigability of the lower extremities on prolonged exercise. Neurological examination was negative.

Comment. A brief résumé of previously reported cases is given in Table 1, which reveals the following facts:

1. Of 25 reported sexes, 18 are male and 7 female.
2. Of 25 reported ages, 18 are 20 years or less.
3. While duration of symptoms is from less than 1 month to 13 years, about $\frac{1}{2}$ had symptoms for 1 year or less.
4. The location of the cyst was mid or low dorsal in all except 5 cases; only 1 was in the cervical region.

SUMMARY

A case of non-traumatic, non-parasitic spinal extradural cyst with patent communication with the spinal subarachnoid space is presented. The complicating factor of fibrosis of the dura in the region of the cyst is noted. A table of recorded cases is included.

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ACTINOMYCOTIC BRAIN ABSCESS

COMPLETE EXCISION WITH RECOVERY

RICHARD C. SCHNEIDER, M.D.,* AND ROBERT W. RAND, M.D.
Department of Surgery, Section of Neurological Surgery, University of Michigan Medical School and Hospital, Ann Arbor, Michigan

(Received for publication August 18, 1948)

In 1878 Israël[9] reported the first case of actinomycosis in man, which had been observed by von Longenbeck 33 years earlier. Ponfick[16] first described actinomycosis of the brain in 1880 when he reported 2 cases. The involvement of the central nervous system by this disease is in itself rare, and there has been considerable discussion as to whether it can be primary. Zeitlin and Lichtenstein[22] stated that actinomycotic lesions of the central nervous system were secondary to a focus elsewhere in the body, and that if no obvious gross primary lesion could be found, the lungs and other organs should be carefully examined after fixation. However, Cope[3] in a monograph on the disease lists 10 cases of isolated involvement of the brain, 6 of which were located either in the third ventricle or near the septum pellucidum and the anterior commissure. Sanford and Voelker,[18] reporting 670 cases of actinomycosis in 1925, and Cope,[3] reviewing 1,330 cases in 1938, grouped the sites of infection into the cervico-facial, thoracic, abdominal, and miscellaneous. The incidence of cerebral distribution was so small that a separate designation was not made, but these cases were merely included in the miscellaneous group of 8 per cent and 5 per cent respectively. In an extensive review of actinomycosis of the head and neck in 1936, Gardiner[6] stated that "actinomycosis of the brain is almost a hopeless situation." The following year Friedman and Levy[5] collected from the literature 108 cases of actinomycotic involvement of the central nervous system; 23 of these were reported as primary. Treatment was symptomatic and the cases were "invariably fatal."

The advent of chemotherapy has changed the prognosis of the disease. Walker[19] reported a cure of the abdominal type of actinomycosis using sulfa drugs. Ogilvie[15] and Dobson, Holman, and Cutting[4] found that these drugs inhibited secondary invaders, thereby permitting the natural body resistance to overcome the actinomycosis. Cures with penicillin were reported in 1944 by Wollgast,[21] Herrell,[8] Christie and Garrod,[2] and by numerous others since that time. Penicillin was not necessarily the only drug used, but it was regarded by all authors as the most important factor in effecting the cure. Kolouch and Peltier[11] reviewed 1,236 cases with regard to the success of therapy in cervico-facial, thoracic, and abdominal cases and concluded that a regime of energetic supportive therapy, chemotherapy, and radical excision of the infected tissue was best. None of the above authors reported on the treatment of actinomycosis affecting the central nervous system. However, in June 1948 Jacobson and Cloward[10] described the cure of a patient with actinomycotic meningitis treated with sulfonamides, penicillin, and streptomycin.

Krönlein[12] in 1910 appears to have been the first surgeon to perform complete excision of a brain abscess. Although not their usual procedure, most neurosurgeons have, in selected cases, completely removed well encapsulated abscesses. Clovis Vincent in 1936 recommended this procedure in practically all cases. In recent years many pyogenic brain abscesses have been treated by radical excision along with

* 10006 Carnegie Ave., Cleveland 6, Ohio.

massive doses of chemotherapy.[1,7,11,14,17,20] LeBeau[13] reported 3 cases of thoracogenic brain abscesses which were cured by these methods.

We are presenting an additional case of apparent cure of an actinomycotic brain abscess, probably of thoracogenic type, which was treated by a similar procedure.

CASE REPORT

On Oct. 13, 1947, V. P., a 45-year-old white male, was awakened from his sleep by a convulsive movement beginning in the left arm, extending to the left leg, and eventually followed

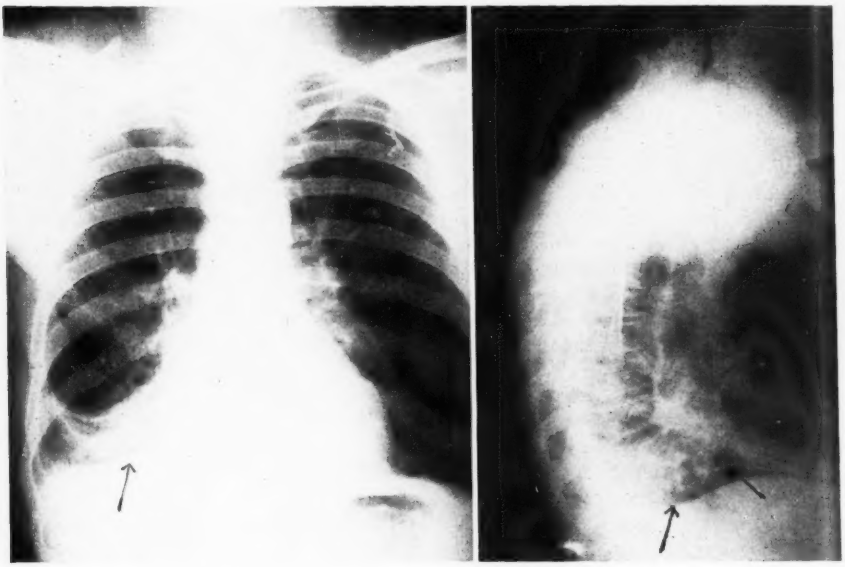


FIG. 1 (*left*). Roentgenogram of chest. Note foreign body (arrow).
FIG. 2 (*right*). Right lateral view showing nodular configuration (arrows) suggestive of neoplasm.

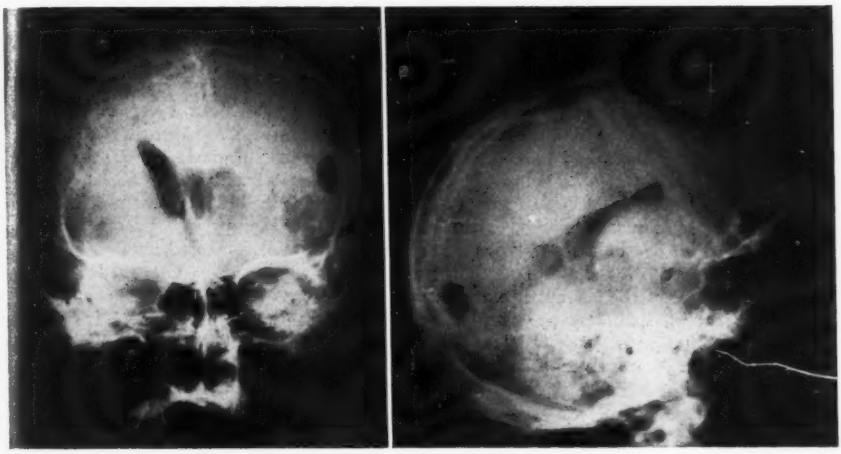
by numbness over the left side of the body. He then lost consciousness, bit his lips, and had sphincter incontinence. Although he was placed on anticonvulsants, he had several similar seizures during the ensuing 10 days before admission to the hospital. Following each episode there was a marked left hemiparesis from which he never completely recovered. The only other significant symptom was vertex headache, which began with the acute phase of his illness.

At the age of 6 the patient had aspirated a piece of leather and a lung abscess developed, which on one occasion caused such severe illness that a fatal outcome was feared. Secondarily there was a chronic productive cough which was noticeably aggravated by an attack of pneumonia in 1940.

On admission to the Neurological Service at University Hospital, the patient's temperature was 99°, pulse 68, and respirations 20. Abnormal neurological findings were limited to slight weakness of the left upper and lower extremities, with hyperactive patellar reflex on the right. There was slight dyspnoea with coarse ronchi throughout the chest and definite moist rales at the bases. The only other unusual physical finding was a generalized shotty lymph-

denopathy in the posterior auricular, inguinal, and epitrochlear nodes. X-rays of the skull showed a calcified pineal gland in the midline, a persistent frontal suture, and no evidence of an intracranial lesion. Films of the chest showed a foreign body (Fig. 1) producing partial secondary collapse of the right lower lobe and a hypostatic pneumonia. The X-ray consultant stated that the nodular configuration in the right lateral view (Fig. 2) suggested a neoplasm. Ophthalmological examination disclosed no papilledema or gross visual field defect.

On Oct. 30, 1947 the patient had a convulsive seizure with flexion of the left arm and rotation of the head toward the left side. A complete left hemiplegia developed and he became



FIGS. 3 and 4. Ventriculograms demonstrating space-occupying lesion in right frontoparietal area.

semi-comatose within a brief period. A diagnosis of right frontoparietal metastatic brain abscess or tumor was made.

Following bronchoscopy with removal of large quantities of tenacious sputum, there was considerable improvement in the respiratory exchange. Ventriculography was performed, demonstrating a space-occupying lesion in the right frontoparietal area (Figs. 3 and 4).

Operation. The dura was extremely tense. A small incision was made in the dura and in the cortex of the premotor area. Upon inserting the exploring needle to a depth of 3.5 cm., an abscess was tapped and several ounces of thick, greenish-gray material exuded, cultures of which were made. The exposure was enlarged. No discrete abscess capsule was found, but upon gentle palpation with the finger, a shotty granular tissue was felt. This was removed completely by suction, making what appeared to be a complete excision of all abnormal tissue and leaving a cavity of about 4.5 cm. in diameter which was in direct communication with the right lateral ventricle. The scalp incision was closed in layers, without drainage.

Postoperatively the patient received 10 gm. sodium sulfadiazene intravenously during the first 24 hours and 5 gm. intravenously daily for 10 days. From that time on he tolerated 1 gm. orally given with an equal dosage of sodium bicarbonate; total dosage was 258 gm. sulfadiazene. He received 40,000 units penicillin intramuscularly every 3 hours and also for the first 4 days 50,000 units intrathecally, making a total of 4,440,000 units penicillin.

The CSF was xanthochromic for a few days after operation but subsequent lumbar punctures showed a colorless fluid with a wbc. ranging from 0–10 lymphocytes. Three cultures were negative for pyogens, and smears of the sputum contained no fungi or tubercle bacilli.

Direct culture from the abscess showed a dispersed "snow flake" growth on Brewer's thioglycolate broth and brain-heart infusion broth. Subcultures to blood agar plates when innoculated in 10 per cent CO_2 showed the typical long, filamentous, branching bacilli with clubbed ends. Colonies were granular and emulsified in physiological saline with difficulty. The organism was not acid-fast. Both colony formation and morphology of the stained organism were typical of actinomycosis. Growth requirements indicated a microaerophilic organism.

Pathological Study. The tissue excised from the brain showed focal accumulations of polymorphonuclear leucocytes, particularly perivascularly, with severe edema and astrocytic proliferation. There was no evidence of granuloma formation in these sections, but only a small portion of the tissue had been preserved. The findings were compatible with a recent brain abscess.

Course. The patient responded well to therapy. Late in his convalescence he was referred to the thoracic surgeons, who agreed that the brain abscess had developed secondarily from the lesion in the right lower lobe. Two bronchoscopies were performed, but the abscess could not be located. Specimens of sputum procured at these times yielded staphylococci and streptococci, but no actinomycosis. Lobectomy was considered, but not performed.

On Dec. 2, 1947 the patient was transferred to the Veterans Hospital at Dearborn, Michigan. He was afebrile but a large amount of sputum was expectorated daily and a complete hemiplegia was still present. An additional 1,920,000 units penicillin were given intramuscularly followed by aerosol penicillin 4 times daily. On this therapy his sputum decreased markedly. Smears and cultures of the sputum were negative for tubercle bacilli and fungi on several occasions. With the aid of physiotherapy his left leg and thigh improved but there was no movement of the left upper extremity.

The patient was examined at home on May 27, 1948. He was mentally alert, but had no recollection of his 6 weeks' stay at University Hospital. There was a left central facial paresis. The left upper and lower extremities were slightly spastic with inability to flex the arm at the elbow or pronate or supinate at the wrist. Extension of the left leg at the knee and movement of the left foot and ankle were absent. With support he was able to walk about 15 yards. There was hyperesthesia over the entire left half of the body with generalized hyperreflexia on that side. Although confined to a wheel-chair, the patient was in good spirits and quite optimistic about the future.

COMMENT

Since the patient had received large doses of chemotherapy and the organism was microaerophilic, it is perhaps not surprising that the fungus could not be grown postoperatively from the spinal fluid or the sputum.

According to Gardiner[6] and Zeitlin and Lichtenstein,[22] the histological changes in the brain due to actinomycosis may not differ from those caused by the ordinary brain abscess. This was true in the case presented above. It is possible that the communication of the abscess with the ventricular system, allowing the intrathecal penicillin to come in direct contact with the contaminated area, was an important factor.

Aerosol penicillin was of great value in diminishing the patient's profuse expectoration of tenacious sputum and may have favorably influenced the primary lung lesion. As long as the pulmonary focus remains, extension to other parts of the body may occur, and therefore a pulmonary lobectomy seems indicated.

SUMMARY

A case of actinomycotic cerebral abscess, most likely thoracogenic, apparently treated successfully by combined total surgical excision and antibiotic therapy, is reported.

The patient received a total dosage of 60 gm. sulfadiazene intravenously during the first 10 postoperative days. The drug was then administered orally, 1 gm. daily, making a total dosage of 258 gm.

Penicillin was administered intrathecally for the first 4 postoperative days in doses of 50,000 units daily. He also received 40,000 units parenterally every 3 hours, making a total dosage of 4,440,000 units penicillin. Following transfer to another hospital an additional 1,920,000 units penicillin were given plus aerosol penicillin 4 times daily.

Pulmonary lobectomy was not performed, but we believe that it should be, to remove, if possible, the suspected primary lesion.

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EXTRADURAL SPINAL CYST

A CASE REPORT*

HANNIBAL HAMLIN, M.D.,† COMDR. RICHARD W. GARRITY (MC), U.S.N., AND LIEUT. (jg) JAMES B. GOLDEN (MC), U.S.N.R.
Neurosurgical Service, U. S. Naval Hospital, Chelsea, Massachusetts

(Received for publication July 29, 1948)

Intraspinal extradural cysts are comparatively rare lesions, although their characteristics have been well described by Elsberg,[4,5] Mixter,[9] Cloward,[2,3] Adelstein,[1] Hyndman,[8] and Hamby.[6] A review of the pathological notes accompanying 22 previous cases in the literature indicates that only 9 were entirely dural in structure and that all but 2 were located in the thoracic division of the neuraxis. The defects were diagnosed and treated in younger individuals, the oldest being 46 years, the average about 3 years.

FIG. 1. Roentgenogram showing localized pedicle erosion (arrows).

The patient described in the present report had led an active life without disability until his condition was demonstrated and ameliorated by surgical repair at the age of 25.

The location of the lesion at a high lumbar level rendered the clinical findings different from those of recorded cases. Moreover, an unusual exaggeration of symptoms by gravitational acceleration was encountered through our subject's occupation as a dive bomber pilot for the U. S. Naval Air Force. The factor of aerial acceleration has been considered as a precipitating cause of lumbar intervertebral disc herniations among aviation personnel. The patient under discussion was studied with this possibility in mind on several different surgical services including our own; and the obvious roentgen sign of localized pedicle erosion (Fig. 1) was overlooked repeatedly.

Unit No. 174871. The patient, a 25-year-old naval aviator, had been admitted to another hospital with a 2-year history of left-sided low back discomfort which had developed gradually

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* The opinions or conclusions contained in this report are those of the authors and are not to be construed as necessarily reflecting the views nor the endorsement of the Navy Department.

† 4 George Street, Providence 6, Rhode Island.

and involved the left gluteal region. Coughing and prolonged standing aggravated the symptoms. Clinical appraisal, including routine roentgenography, produced a diagnosis of arthritis. He returned to flying and carried on for 11 months before again seeking medical advice. During this time he noticed that in pull-outs from dives, he would lose control of his leg for a short time followed by a tremor which lasted a few moments longer. He also complained of pain on turning the head or trunk to either side. This discomfort was referred to the low back.

He was then admitted to another hospital and transferred to the Chelsea Naval Hospital for further study. His complaints on admission in late Jan. 1947 were obscure discomfort in the back, loss of spring in the legs and some slight gluteal pain on coughing.

Examination. The physical and laboratory findings were normal and roentgenograms were reported as negative. CSF pressure was normal as were dynamics, cell count, Wassermann, protein and gold sol test. Subsequent study by the general, orthopedic, urologic and roentgen services arrived at no diagnosis and he was referred to the neurosurgical service. The only abnormal finding at this time was a CSF protein of 231 mg. per cent. Lower lumbar myelogram proved to be normal, but a CSF sample taken concurrently revealed a protein of 531 mg. Also at this time both Achilles reflexes were missing with reinforcement. The dynamics remained normal. Simultaneous cisternal and lower lumbar taps then revealed protein of 25 and 745 mg. per cent respectively. An immediate repeat myelogram disclosed an abnormality at the 1st lumbar interspace. Stereoscopic views demonstrated an almost complete extradural block at the lower margin of L1. By this time some sacral hypalgesia and hypesthesia had developed.

FIG. 2. Gross appearance of extradural spinal cyst at operation

Operation. On May 29, 1947 a laminectomy centered over the 1st lumbar segment revealed a definite dorsal displacement of the lamina, and removal exposed a bluish cylindrical structure believed to be dura. Incision of this membrane was followed by a gush of crystal clear fluid but, as in Hyndman's case, we were greatly surprised to see no evidence of conus or cauda equina. Extension of the laminectomy to the lower margin of the 11th thoracic segment demonstrated the abnormal structure to come to a pointed juncture at the right T11–T12 interspace. It was attached in a broad flat expanse to the ventral aspect of the upper portion of the 2nd lumbar vertebra. Two pouches also presented on the left which bulged into the T12–L1 and L1–2 foramina. Elevation of the cyst visualized a normal pulsating dura beneath. The cystic mass was entirely emptied of fluid prior to the attempt to interrupt its upper attachment. However at this point a nerve root was seen and, on separation, a spinal fluid leak occurred which ceased spontaneously within a few moments. The CSF was thought to come from the subarachnoid space. [This may have accounted for the small area of numbness near the upper end of the incision which the patient experienced during the following year or more.]

Postoperative Course. A small cystic mass developed beneath the incision which disappeared after 2 weeks of pressure dressings. Because of the prevalence of kyphosis (Elsberg[4,5]) in these cases, the patient was fitted with a steel-ribbed corset which he has worn to the present time without the appearance of back deformity.

Over a year later he wrote from a midwestern city, where he is gainfully employed, that his troubles are really very slight, that he has some pain in the right leg on heavy lifting, some numbness about the upper end of the incision on the right side with some gurgling sensation; also, headaches which are immediately relieved by lying down.

COMMENT

This report serves to emphasize the importance of a careful roentgen study of the vertebral pedicles and arches (Elsberg[4]); and again stresses the possibility of error in the commonly used tests for manometric block. Not infrequently useless operations have been done for suspected conditions actually caused by lesions of the cord

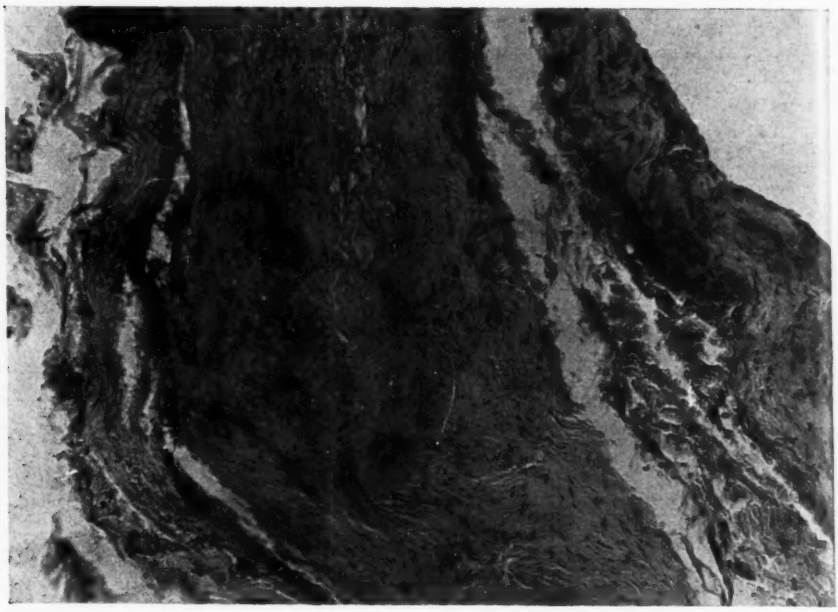

FIG. 3. Photomicrograph of cyst showing resemblance to dura.

or its appendages at higher levels. It is thus apparent that the safe procedure is to carry myelographic studies to several segments above the highest somatic sign.

Since the majority of the reported lesions have been thoracic, the location in this instance, associated with the late loss of Achilles reflexes, suggests that the neurologic signs may be variable and that spasticity need not be present, although the "loss of spring" in this case may have been due to transient subclinical alteration in muscle tone.

This cyst (Fig. 2) does not appear to represent either the traumatic or the postoperative variety described by Swanson and Fincher,[11] nor does it resemble those described by Tarlov[12] or Rexed[10] in which the cysts were small and continuous with the arachnoid and dura. As may be noted in Fig. 3, the lesion resembles dura. It lacks the characteristics of the intradural type described by Haymaker and Foster.[7] The tendency of the cyst wall to evaginate into the intervertebral foramina causes speculation on the possibility that this condition, if allowed to progress, might develop into meningocele-like reservoirs which would tend to decompress the cord.

Although the exact frequency is unknown, a random summation of 5 large series

comprising lesions of the spinal cord or its coverings, exclusive of herniated discs, revealed only 2 extradural cysts. During the war years 1940–1945, there was not a single case reported from the hospitals serving the Navy, Marine Corps, and all their dependents (Ware[13]).

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A NEW TYPE OF CERVICAL BRACE*

Charles Spicer, M.D., and Captain Allan E. Bayless, M.C., A.U.S.

Department of Neurosurgery, Birmingham Veterans Administration Hospital, Van Nuys, California

(Received for publication June 14, 1948)

A new modification of cervical bracing has been developed by our Neurosurgical Department in conjunction with technicians of the hospital Brace Shop.† The cervical brace is a simplified modification of the Thomas cervical brace. It is extremely light and can be manufactured easily and economically.

The brace (Fig. 1) consists of a chin rest and occipital rest which are connected with single light metal strips to the chest and back pads. The chest and back pads are supported by single adjustable canvas straps which ride over each shoulder. The

* Published with permission of the Chief Medical Director, Department of Medicine and Surgery, Veterans Administration, who assumes no responsibility for the opinions expressed or conclusions drawn by the authors.

† The technicians to whom we are indebted are Mr. John C. Russey, Mr. John C. Green, and Mr. Eugene N. Capaiu.

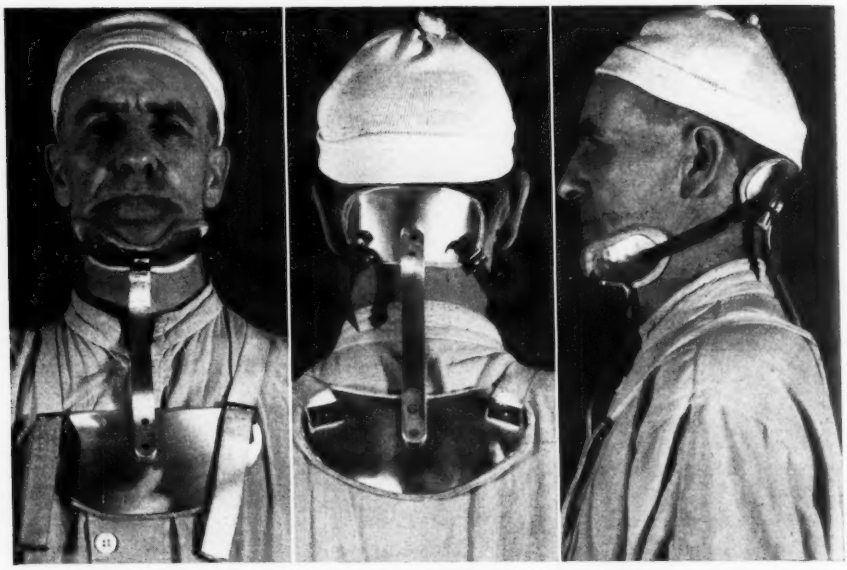

FIG. 1

chin and occipital rests are connected by adjustable leather straps. Another modification is found in the chin rest, which is cut away over the symphysis of the mandible, thus eliminating an uncomfortable pressure point. The pressure of the chin rest is distributed along the inferior margin of the mandible.

The degree of traction pressure is regulated by the degree of tightness of the shoulder straps. The position of the adjustable leather straps allows for ease in adjustment.

The braces have been manufactured from surplus aluminum alloy and all parts are easily bent to fit the variations in individual anatomy. The chin and occipital rests as well as the chest and back pads may be cut from patterns for mass production and then bent to fit the individual requirements. The parts are simply riveted together and the rests and pads are lined with inexpensive felt. These may also be covered with soft leather as desired for a more finished product.

Such a brace is light but still offers adequate comfortable traction. Also due to its simple design the brace may be produced in large volume economically with a relatively short assembling time.

Approximately 200 such braces are at present in use by our service, and they have been found to be satisfactory for the treatment of the average patient requiring cervical bracing.

Obituary

MAX MINOR PEET

1885–1949

The death of Max Minor Peet on March 25, 1949 was a rude shock to his many friends. He had felt ill that morning, but in spite of this he did not shorten his eight o'clock lecture to the Junior students. Death came suddenly an hour later from coronary thrombosis.

Doctor Peet was such a youthful, vital person that it did not seem possible that he was sixty-three. He was born in Iosco, Michigan, October 20, 1885 and was the son of Lafayette and Eunice Ann Peet. He received his A.B. degree from the University of Michigan in 1908 and an M.A. and M.D. degree in 1910.

He early showed promise in the field of science while attending high school in Ypsilanti. An ornithology contest for college students was being held at the nearby University of Michigan. The budding ornithologist asked if a high school student might enter the competition, and upon receiving permission, proceeded to win first prize. This was an original and very beautiful Audubon print.

Doctor Peet went on to win world fame as an ornithologist. His first scientific publication written in 1905 was entitled "Observations on the Nesting Habits of a Pair of House Wrens." He started collecting bird specimens early. As late as 1932 he was sent by the University to the Chisos Mountains in Texas to collect. He was a crack wing shot, as was his father before him, and he succeeded in bringing back a number of specimens of the White Throated Swift, the fastest flying bird in the United States.

He claimed that any proficiency that he acquired in surgery started with the preparation of Hummingbird skins, a most difficult technical procedure. The specimens he prepared with his own hands were second to none. His ability to prepare specimens of day-old nesting House Wrens, Blackbirds, and the like was probably unique. The delicacy of the baby birds makes them exceedingly difficult to skin. Probably no one else ever mastered this particular technique so perfectly.

Doctor Peet was one of the early discoverers of the nesting places of the rare Kirtland Warbler. From a description given him by a rural mail carrier he believed that this bird, which nests only in Michigan, had been seen near Roscommon. While he was driving in a horse-drawn buggy he heard a song he had never heard before. He thought that this must be the Kirtland Warbler, which proved to be the case. Some of the specimens he took at this time are in the Smithsonian Institute and the American Museum of Natural History.

His own collection of birds numbered approximately 15,000 and was probably the largest private collection in the country. He was interested in obtaining a completely representative collection of American birds found north of the Rio Grande; this entails 1400 species and sub-species. There still remained a few gaps to be filled and he was constantly in touch with field ornithologists to fill these. There was scarcely an ornithologist in the country who had not at one time or another borrowed from the Peet Collection. At the time of his death he was engaged in an x-ray study of the skulls of the two kinds of Golden-eye Duck in an attempt to identify these birds finally, on a basis of skull structure rather than plumage. This original method had already created a great deal of interest though it had not yet been published.

In spite of his active interest in Natural History both before and after entering medical school, he had always planned to become a surgeon. His surgical career began with an internship at Rhode Island General Hospital. After two years he went to the University of Pennsylvania as Robert Robinson Porter Fellow in Research Medicine. He was assistant chief surgeon at the Philadelphia General Hospital from 1914 to 1916. At this time he did a number of experimental Eck fistulae. With Doctor Charles H. Frazier, by whom he was trained, he probably attempted the first Eck fistula on the human.

In 1916 he returned to the University of Michigan as Instructor in Surgery. What little neurosurgery there was, he did. It was only later, however, when Doctor Hugh Cabot came on as Professor of Surgery in 1919 that he began to specialize. He continued to do some general surgery until 1926, when he reluctantly gave it up. He became Professor of Surgery in charge of the Division of Neurosurgery in 1930.

In 1925 he introduced hypertonic glucose for the control of increased intracranial

pressure. In 1926 he published the first large series of antero-lateral cordotomies. He was the first to operate for subdural hematoma in infants.

He early became famous for his technique in the operation for tic douloureux which he had learned from Doctor Frazier. About 1930 a group of plastic surgeons met in Ann Arbor. Some of them went in to watch Doctor Peet operate for tic douloureux by the temporal route. Doctor Vilray Blair, unknown to the operator, timed him with a stop watch—seven minutes from the skin incision to exposure of the sensory root. He did over a thousand of these operations during his lifetime. He once did four of them between 9:00 A.M. and 1:00 P.M., using the same table and local anesthesia in each case. It has been said that Dandy was the greatest expert on the surgery of the posterior fossa. The same might be said of Peet for the middle fossa.

Doctor Peet was best known for his work on hypertension. The evolution of the operation here is an interesting story. A patient with the gastric crises of tabes had failed to respond to cordotomy. It was thought that cutting the splanchnic nerves might relieve the pain. The operation was worked out on a cadaver. In February, 1929 the splanchnic nerves were divided and part of the dorsal sympathetic chain excised. The procedure was carried out bilaterally through an extra-pleural, supra-diaphragmatic approach. The operation was unsuccessful but paved the way for later work. The operation had been performed only once before, as far as is known, by Otfrid Foerster of Breslau, also for gastric crises. It escaped notice as it was only mentioned in a book on pain written in German. Pieri had published a similar anatomical approach in 1927 but he did not perform the operation until June, 1929 when he performed a unilateral splanchnic section for gastric atony.

The first splanchnic section for hypertension at the University Hospital was done in November, 1933. The patient had been sent to Doctor Peet as probably suffering from brain tumor because of the presence of four diopters of papilledema and rapidly failing vision. His blood pressure on entrance was 270/150. It was only after a period of study that it became certain that this was not a brain tumor, since the syndrome of malignant hypertension was not clearly delineated at this time. The operation was fortunately a success and the patient lived a normal existence for over ten years. He performed this operation bilaterally over eighteen hundred times.

Doctor Peet was a member of many societies. These included the American Neurological, the American Surgical Association, and the Harvey Cushing Society. He was an honorary member of many others. His favorite society, however, was the Society of Neurological Surgeons to which he was elected in 1922 and of which he was president in 1935. He was rarely absent from one of their meetings. He was intensely loyal to his friends in this small society, and was proud of their achievements.

At the time of his death he was chairman of the American Board of Neurological Surgery. He had been a member of the general advisory committee of the National Foundation for Infantile Paralysis for some years. He participated in the work of the National Foundation from its inception. It was a striking coincidence that because of his interest in infantile paralysis it was he who obtained the material from which the Lansing strain of poliomyelitis was isolated. This was the first strain that was shown to be adaptable to cotton rats and mice by Doctor Charles B. Armstrong of the National Institute of Health.

Doctor Peet married Grace Stewart Tait in 1915. Their devotion to each other and to their children was a wonderful thing to see. Many neurosurgeons from here and abroad knew the disarming hospitality of their household.

Perhaps the outstanding characteristic of Doctor Peet was his informality and infectious good humor. Affectation was left out of his make-up. He was so secure in himself that he did not know the meaning of jealousy.

An excerpt from a letter received from a girl he had operated upon some years before sums up the man: "I have always wondered at his kindnesses down the years and have felt it a privilege to be even remembered. I shall never forget his always having time to see me and listen, nor his interest in my husband and child. Of the many contributions he made, not the least was his graciousness and kindness to the little people."

Edgar A. Kahn, M.D.

CARIES CRANII

(TUBERCULOSIS OF THE FLAT BONES OF THE VAULT OF THE SKULL)

A. C. DE VET, M.D.

Neurological and Neurosurgical Clinic "St. Ursula," Wassenaar, Holland

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THREE reasons seem to justify the report of the following case of caries cranii. (1) The disease is an uncommon condition to which little attention has been paid by neurosurgeons. (2) Although rare, neurological symptoms may be caused by tuberculosis of the skull. These symptoms were among the outstanding features in our case. The disease therefore is one with which neurosurgeons should be familiar. (3) Streptomycin was used locally and the possible influence of it is discussed.

It is difficult to find a full description of caries cranii in the neurosurgical literature. Dandy[4] merely mentions the disease, states that the sick bone has to be removed completely, and gives no results. Davis[5] also refers to it very briefly and probably quotes from the article of Strauss[18,19] (see below). Obviously, Sachs[14] and Bailey,[2] whose books deal only with cerebral tumours, also not mention it, but that the subject is not discussed by Maxwell[10] in his description of congenital traumatic and inflammatory disorders of the cranium, indicates that neurosurgeons generally have no great interest in tuberculous infections of the skull.

Textbooks of surgery as a rule give little attention to the disease: Dandy[4] has been cited above. Gauvain[7] speaks of a rare but serious condition which starts in the diploe but results in general dissemination with correspondingly bad prognosis. This statement is not in accordance with the experiences reported by Strauss,[18,19] Joutard (cited by Strauss) and others, but Babcock[1] holds the same opinion, giving a mortality rate of 50 per cent.

Pancoast, Pendergrass and Schaeffer[12] in their textbook on roentgen diagnosis give the impression that among radiologists also there is only scanty interest. Even in books on tuberculosis of bones and joints, there may be no mention whatever of caries cranii (Girdlestone[8]), or it is discussed only briefly (Kremer and Wiese,[9] Sorrel and Sorrel-Dejerine[16]).

Although I can by no means be complete in reviewing books, the above citations from well-known examples are sufficient to show how little information they contain on this subject. Those who are particularly interested will have to resort to special articles. However, as far as I could trace, these are exceedingly rare in neurosurgical literature; neither in the *Zentralblatt für Neurochirurgie* nor in the *Journal of Neurosurgery* has any article on caries cranii been published.

One of the most important and recent articles is that of Strauss,[19] an excellent review, in which he collected 200 cases from the current literature and added 3 of his own. Many of the details, mentioned below, are taken from this review.

It is generally accepted that the tuberculous infection reaches the bone of the skull through the blood stream almost always from a distant focus. Several cases of so-called primary infection are reported but only in those where an open wound has been contaminated or the infection is directly conducted from an existing tuberculous infection in other parts of the head (ear, nose, eye, etc.) can this be fully accepted; in all other cases there remains some doubt about a concealed tuberculous focus elsewhere in the body (thoracic or abdominal lymph nodes). In the majority of cases there was phthisis of the lung or tuberculous cervical lymphadenitis, but many other localisations of tuberculous processes have been reported as the supposed primary source of the cranial infection.

The haematogenous spread makes it acceptable that the initial localisation is in the cancellous bone of the diploe. Because the flat bones of the skull contain such a small quantity of diploe it is easily understood that the number of skull infections is very low compared with that of other bones. Of all cases of bone tuberculosis in different statistics the localisation in the skull was present only in 1.37 per cent (Parel), 1.25 per cent (Claeys), and 0.2 per cent (Berck). As the frontal and parietal bones contain relatively more diploe than the other flat bones of the vault, it is a logical consequence that these two bones (which, as another argument, together take up about three-quarters of the surface of the skull!) show the highest incidence of infection (172 out of 206 cases). It may be that the influence of trauma, which according to Schüller (1878), Volkmann,[20] Strauss[19] and others is indisputable in many cases, has to be taken into account too, because the frontal and parietal bones are more exposed to trauma than the other parts of the skull. It is generally observed that children show a much higher tendency to caries cranii than people in the later decades of life. Three-fourths of all cases occur before the 20th year (Sorrel and Sorrel-Dejerine[16]). A satisfactory explanation cannot be given. Strauss,[18] Reber, and others discuss the possible influence of the numerous traumata in childhood; the fact that males are more often afflicted than females (Labhardt's statistics showed 69 per cent in males and 31 per cent in females), which is ascribed to the rôle of a trauma, they regard as a point in favour of their supposition.

Most authors agree in distinguishing two types of the disease: *a*, the circumscribed (perforating) type (Volkmann,[20] Gangolphe), and *b*, the diffuse progressive type (Koenig).

Type a generally is described as a round, usually punched-out looking defect through the entire thickness of the skull. This can be seen *in nature* as well as in the roentgenograms. However, if one pays more attention to the various differences in the spread of the infection throughout the bone, it would not be surprising if atypical defects would prove to be more numerous.

In the majority of cases it is stated that the tabula interna is more widely affected than the external table; sometimes only the diploe are involved. These various conditions must give different X-ray pictures and in our own case one cannot speak of really sharply outlined round defects. The centr

shows the highest degree of decalcification; this gradually decreases toward the periphery (Figs. 1 and 2). This corresponds with the operative findings (see later).

Volkmann[20] believed that the perforating type is always limited to a single area but the existence of two (rarely more) separate areas is so often observed, including our own case, that this statement is not valid. The hematogenous spread of the infection readily accounts for multiple foci and it is curious that type *b* is so much less frequent than type *a*.

In *type b* there is a widespread infection of the cancellous bone and most often of the inner table too. But the tabula externa shows only a number of perforations which seem to be separate. The findings in type *b* have cast some doubt whether in type *a* a real independence of the two (or more) areas existed. St. John goes so far as to deny this independence and he does not approve of distinguishing two different types.

However, the operative findings in the case of Strauss[18] and in our own case, as well as in many others, have given proof of the existence of complete independence of two or more areas. Moreover, the clinical course of type *a* is quite different from that of type *b*.

The perforating type develops slowly, and the patients come under observation because of the swelling on their head. This is often combined with, and sometimes preceded by, headache. If an X-ray is taken in this stage, the perforation of the skull in most instances is already present. The swelling can be ascribed to an abscess which first remains subperiostally but afterwards breaks through this layer and affects the skin until a sinus develops. Sometimes, however, the swelling consists mainly or only of granulations. The whole course takes many weeks or months and sometimes the infection stops and the symptoms subside before a fistula is formed (Babcock[1]).

In contrast to the perforating type, the progressive form takes a rapid course and spreads over an unlimited area of the skull. The operative treatment is difficult, not only because big areas of the skull have to be removed (and even then the limits of the infection cannot always be reached), but also because of the extensive tuberculous granulations which develop on the dura mater. The prognosis in these cases seems to be much less favourable, although definite figures are not given.

The dural granulations are present as soon as the infection perforates the inner table. In type *a* they originally remain restricted to the area of dura facing this perforation of the inner table, but after some time they spread over a small circular area in the vicinity, generally not exceeding a few cm. Strauss[19] states that, in his own case as well as in those of other authors, these granulations are not very adherent to the dura and that they could be stripped off easily. My own findings are not consistent with this statement. In the centre of both areas the granulations were absolutely fixed to the dura and parts of them had to be left there because of the danger of perforation of the dura.

The granulations themselves do not penetrate the dura. Tuberculous

meningitis therefore is a rare complication; it has been reported in only 10 out of 223 cases of caries cranii. Cerebral tuberculosis is even rarer (5 cases). Although granulations are present almost without exception, it is unusual to find abscess formation between dura and skull and if so, the collection of pus is very small. Outside the skull, however, under the periosteum, afterwards under the muscles and the skin, one or more abscesses may be present; they infiltrate the muscles and the skin until they find an outlet which discharges for long periods if no treatment is installed.

"In a few cases the granulations have formed a large, tumour-like mass (in one case the size of an orange) and then they may give rise to pressure symptoms like a tumour." This is the only comment Strauss[19] gives about a detail which is of primary interest to the neurosurgeon, without reference to any author. The same reserve is to be found when he describes the general symptoms which may accompany caries cranii without granulomatous tumour formation: "signs of cortical irritation with epileptic seizures have been observed in a few cases." It therefore seems justified to report more fully about the cerebral disorders met with in our own case.

Case History. C. L. Th., male, 20 years old.

The first signs of the patient's illness developed acutely on June 27, 1945. He complained of pain on the right side of his chest and his temperature was raised. A pleurisy was diagnosed and he was kept in bed until March 1946. After careful mobilisation he showed signs of renewed activity and sanatorium treatment was instituted. At the time of his admission (Sept. 1946) he knocked his left forehead against a door and on that spot a lump developed in the course of a fortnight. This lump remained present but varied in consistency; sometimes it was soft, at other times it was rather hard. It was soon followed by a second lump in the left parietal region (without a preceding trauma). This lump increased progressively in all dimensions whereas the first one was more constant in size. Often the patient complained of local headache near both lumps and of local tenderness on pressure. In mid Oct. 1946 neurologic symptoms complicated the clinical picture. A sensation of numbness developed in his right arm, soon followed by the same sensation in his right leg and the right half of his face. In November he had a spell of unconsciousness, lasting several hours. There were no epileptic manifestations at that time. This was followed by some weakness in the right half of his body. In the last week of Dec. 1946 he had clonic contractions in the right arm and fingers, and they occurred several times after that. Never were there any contractions in his face or in his leg.

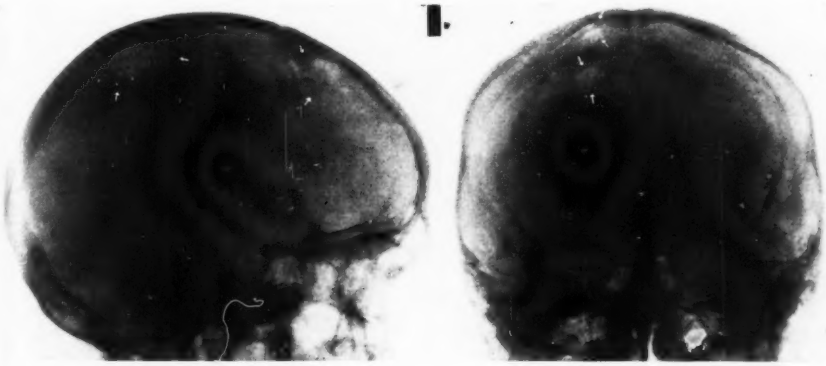
On Jan. 1, 1947, he was transferred from the sanatorium to our clinic. During the last months his headache had slightly increased. In the sanatorium no tuberculous changes had ever been found in the lungs nor elsewhere in the body, but the pleurisy was always considered as of tuberculous origin, as were the lesions in the skull.

Examination. In the left frontal and parietal regions an area of decalcification was clearly visible in the roentgenograms (Figs. 1 and 2). His right-sided pleurisy was considered to be still active because sedimentation rate was 15 mm. after 1 hour, 24 mm. after 2 hours (Westergren). His temperature was subfebrile (max. 37.7 in the evening).

Neurological examination revealed a slight right-sided hemiparesis of the arm and face and a hardly noticeable one of the leg. All qualities of sensibility were

slightly diminished. The fundus oculi showed no papilloedema but at lumbar puncture CSF pressure was raised (190 mm. in horizontal position). The CSF composition was hardly abnormal; cell count, normal (1/3 per mm.^{3}); total protein, 41 mg. per cent; protein quotient, 0.7; serological reactions negative. Colloidal gold reaction of Lange, 1223221100; colloidal benzoic reaction of Guillain, 000000555500000; mastix reaction of Emanuel-Kafka, completely normal.

After a short period of observation it was evident that the focus in the parietal region was active and that the patient's condition had deteriorated. A puncture in the parietal lump yielded a negative result: no pus. In order to establish the nature of the process, a small excision was made for pathological-anatomical examination. The tissue showed typical tuberculous changes. It was curious, however, that with



FIGS. 1 and 2. Lateral and A.P. roentgenograms of skull showing frontal and parietal tuberculous areas on left side.

the Ziehl-Neelsen stain no tubercle bacillus could be seen and that the Löwenstein cultures remained negative. Because of the progressive neurological picture it was judged necessary, and because of the absence of meningeal irritation it was expected to be sufficiently safe, to remove both of the tuberculous cranial foci.

Operation, Jan. 29, 1947. Frontoparietal curve-linear incision. The skin flap was dissected from the periosteum and turned up in temporal direction.

Granulomatous tissue was found in the frontal and parietal regions, where it was only slightly connected with galea and periosteum. It invaded the skull, which was partly perforated (Fig. 3); in other places only the inner or outer table of the skull was eroded. The granulomatous tissue was in very intimate contact with the dura, especially where the bone had disappeared.

All pathological bone was nibbled away until sound-looking bone appeared all around, and then normal bone was also removed, until a margin of $\pm$ 1cm. of normal-looking dura was laid bare. It was possible to remove most of the granulomatous tissue from the dura, but in the centre of both areas the connection was so intimate that the risk of perforating the dura was too great, and a thin layer, probably containing granulomatous tissue, had to be left. In the thickest parts the granulomatous

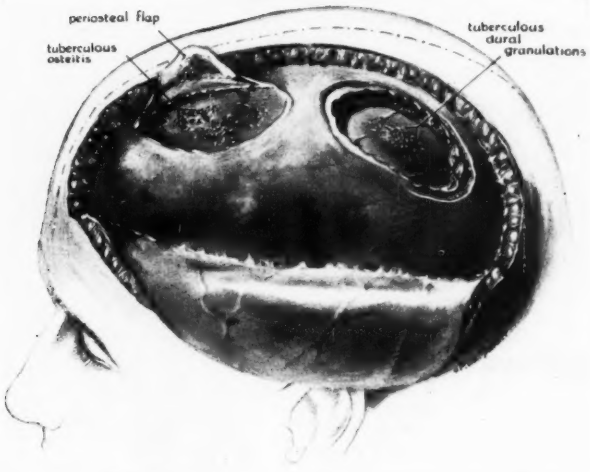


FIG. 3. Sketch of tuberculous areas as seen at operation, the frontal area before, the parietal area after the diseased bone had been nibbled away.

tissue measured 1½–2 cm. The diameters of the remaining bone defects were: frontal, 5×6 cm.; parietal, 7×7 cm. (Fig. 4).

After haemostasis, a solution of streptomycin (1 gm. in 20 cc. physiological salt solution) was spread over the whole operative field in order to diminish the risk of dissemination of the tuberculous infection. Subsequently penicillin-sulphamezathin powder was used to prevent secondary infection. Finally the wound was closed in one layer with separate catgut stitches.

The *postoperative course* was remarkably uneventful. The neurological signs disappeared within 7–10 days. The wound healed without any discharge. The temperature returned to normal within 6 days, but during the first fortnight it occasionally rose to 37.8 or 37.9 in the evening. After a fortnight, however, it remained completely normal, not exceeding 37.4.

FIG. 4. Frontal and parietal bone defect, one year after operation.

On Mar. 21 he was sent back to the sanatorium. His sedimentation rate at that time was 9 mm. (Westergren), afterwards reduced to 6 mm. On Nov. 1, 1947 he was sent home from the sanatorium, without any sign or symptom of activity of his tuberculous focus.

On April 16, 1948 he came back because he wanted both his skull defects to be closed; probably this will be done without much delay.

DISCUSSION

Attention may first be called to a few noteworthy details. It is interesting, for instance, that the first area developed so shortly after a trauma that the connection with this trauma was accepted by the patient as well as by ourselves long before we knew from the literature that in many cases a similar role of a trauma had been described.

The second area developed without a preceding trauma. Never could any direct anatomical connection between the two areas be demonstrated, neither in the roentgenograms nor during the operation. It is probable, therefore, that both areas originated from the same haematogenous spread but that at the first one, at the spot of the recent trauma, developed more quickly because this area was a *locus minoris resistentiae*.

Before the operation there was a constant slight rise in temperature and the sedimentation rate was increased; these were thought to be a consequence of a still active pleurisy. A fortnight after the operation, however, the temperature was completely normal and the sedimentation rate was gradually decreasing. It is improbable that an active, chronic pleurisy was turned into a sudden recovery by an operation on the skull; it is more likely that both the subfebrile temperature and the increased sedimentation rate were caused by the caries cranii, and it is worth while to pay attention to these symptoms in other cases of this disease. However, in most instances it will be difficult to exclude the influence of other tuberculous processes in the body.

The most difficult differential diagnosis is versus syphilis. Not only may the clinical courses be completely similar, but the neurological signs, the X-ray pictures and even the pathological-anatomical findings may be identical. In our case, too, it was not possible to be absolutely sure about the diagnosis on the basis of the microscopic specimen only. It was on other grounds (negative Wassermann in blood and cerebrospinal fluid, the presence of another tuberculous infection in the body, and other details of less importance) that the diagnosis was considered beyond doubt, in spite of the fact that no tubercle bacillus had been found with the Ziehl-Neelsen stain and that the Löwenstein cultures remained negative. Inoculation of a guinea-pig was not performed because at that time there was not the slightest doubt as to the nature of the disease; afterwards we realized that it would have been better to have made use of this additional diagnostic procedure.

Other affections of the skull that can more or less resemble caries cranii are: acute or chronic osteomyelitis; primary destructive tumours of the skull or of the underlying meninges; less frequently, a glioma with erosion of the skull; and finally a number of rarities (actinomycosis, angioma, osteitis fibrosa cystica generalisata, hydatid cyst, and even cephalohaematoma).

The diagnosis of caries cranii, before biopsy or operation, has to be based on the following signs and symptoms: Swelling (single or multiple) of the head, sometimes combined with local headache and tenderness on pressure;

in later stages the swelling may discharge through a fistula. Neurologic symptoms may be present but are rare. In the roentgenogram one or more perforations of the skull are visible; generally they are sharply outlined but vague contours also can exist. A positive Mantoux or von Pirquet test, subfebrile temperature and increased sedimentation rate justify the supposition of a tuberculous nature, while the history of a previous tuberculous process elsewhere in the body (or its discovery during examination) is even more decisive. Puncture, aspiration, smears, cultures and a guinea-pig inoculation can convert a supposition into certainty.

The prognosis of caries cranii in itself seems to be rather good, especially in the circumscribed type. The progressive type cannot always be stopped in its development by surgical interference, as is usually the case in the other type. Strauss[19] is optimistic in his prognosis and gives the following statistics in support of his view. In 87 operations in the literature (76 from Pelletier,[13] 9 from Joutard and 2 from Strauss[19]) 60 complete cures were obtained. There were 9 incomplete cures, that is with fistulas, and 18 patients died. We can now add our case of complete cure to this list.

Other authors, however, give a more grim picture. Gauvain[7] speaks of a serious condition and states that the prognosis is bad because of general dissemination; perhaps his cases belonged mainly to type *b*. Babcock[1] reports a mortality of 50 per cent.

The ultimate outcome for the patient, however, depends largely on the seriousness of associated tuberculous lesions elsewhere in the body. Ménard and Bufnoir,[11] as well as Sorrel and Sorrel-Dejerine,[16] state that cranial tuberculosis is very often associated with advanced tuberculosis of other organs. These lesions not only endanger life but, because they bring the general condition of the patient to a lower scale, the general and local resistance against a spread of the tuberculous area of the skull is diminished.

The treatment from the beginning has been radical surgical removal of all diseased tissue. Babcock,[1] however, speaks only of nutritional measures, radiant, and injections of tuberculin, and Sorrel and Sorrel-Dejerine[16] report several cases in which recovery occurred without intervention, even after many years of fistulous discharge.

There are two types of incision. The German technique, preferred by Strauss,[18,19] uses a straight incision over the affected area; the underlying diseased tissues are removed as radically as possible, including the dural granulations. Afterwards the dura and bone edges are swabbed off with antiseptics (tincture of iodine, iodoform powder, zinc chloride, or others).

The French advise making a horseshoe-shaped incision, wide off the cold abscess, and turning down all the soft tissues so as to expose the diseased bone. The soft parts have to be removed as far as they are infected.

The latter technique was used in our case because it seemed to be of some advantage to have the healing wound away from the infected area and also because by this method it was easily possible to attack both areas in a single session.

The local use of streptomycin was more or less a substitute for systemic treatment. At that time there was not enough streptomycin available for a long-continued intramuscular treatment and it was also doubtful whether such an expensive treatment was justified. Because of the excellent results with local treatment of brain abscesses with a single injection of penicillin it was thought advantageous to give the patient the chance of a possible beneficial influence of local streptomycin in a high concentration. Only recently this local application has received some scientific support. Smith and Waksman[15] have reported on the tuberculocidal properties of streptomycin which were, apart from other factors, dependent upon the concentration, and Garrod[6] gave evidence that streptomycin, contrary to penicillin, multiplies its activity with increasing concentration.

The fact that one patient recovered promptly after local application of streptomycin has almost no value; the single case proves only that streptomycin in strong concentration has no harmful effects on the tissue. This justifies the use of it in other patients and even seems to indicate that streptomycin should be tried locally by injecting it into the infected area, eventually repeating the injection several times. It may be that by this method of treatment surgical interference can be avoided in cases not too far advanced and that, in the progressive type of caries cranii, the tendency to spread rapidly can be checked.

In connection with the above-mentioned experience, attention may be called to two other observations. The first is that of Chauvet,[3] who reported the good influence of local use of streptomycin when it was introduced in the tuberculous cavity of patients treated with Monaldi's drain. The second is an observation in our clinic; because of severe headache and papilloedema a subtemporal decompression was performed on a patient with tuberculosis of the right lung and spine as well as multiple cerebral tuberculomas. The exposed area of the brain showed several small cortical and subcortical tuberculomas (the size of a pea). Because there was almost no streptomycin available, we knew that the patient could not be extensively treated with it after operation. It therefore was decided to test the local action of streptomycin on the tuberculomas; 4 cc. (50 mg. per cc.) were injected under the pia mater in 3 of the cortical tuberculomas and 1 subcortical lesion, surrounding and infiltrating them as much as possible.

It was $3\frac{1}{2}$ months before the patient died. Multiple tuberculomas were scattered throughout the brain. The microscopical picture of the injected tuberculomas was to a certain extent similar to that of the non-treated ones. The necrosis and fibrinisation, however, were unusually marked, whereas proliferation was almost absent. Corresponding differences in microscopical structure were reported, although not fully comparable, in the study of Straub and Schornagel[17] on the aspect of tuberculosis after treatment with streptomycin. These facts give some reason to assume that streptomycin, locally applied, may have some influence on tuberculous tissue. Further observations, however, are necessary to confirm this supposition.

SUMMARY

Description of a case of tuberculosis of the left frontal and parietal bones of the skull (caries cranii). The diagnostic problems and the therapeutical procedures, including the local application of streptomycin, are discussed.

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DESCRIPTION OF A SKIN GALVANOMETER THAT GIVES A GRAPHIC RECORD OF ACTIVITY IN THE SYMPATHETIC NERVOUS SYSTEM*

CURT P. RICHTER, PH.D, AND FREDERICK G. WHELAN, D.E.
Psychobiological Laboratory, Phipps Clinic, Johns Hopkins Hospital, Baltimore, Maryland

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THIS paper contains a description of a skin galvanometer that we have used for over 8 years to record and study activity within the sympathetic nervous system of psychiatric and neurosurgical patients and of normal individuals. It records a phenomenon that was first described by Féré in 1888 and by Veraguth and Müller in 1904 (Veraguth[6]). It has since been become known either as the psychogalvanic or more simply as the galvanic skin response. Féré showed that in patients with hysteria a variety of excitations—visual, auditory, gustatory, olfactory, and emotional—decrease the resistance that the body offers to the passage of a minute direct electrical current. He assumed that normal individuals would show much the same response.

It has been demonstrated (1) that the changes in resistance are localized in the skin, not in the rest of the body (Farmer and Chambers,[2] Richter[4,5]), (2) that this phenomenon depends on the intactness of the sympathetic nervous system (Wang and Richter[7]), and (3) that it has a latent period of 1 to 2 seconds (Veraguth[6]).

Although many workers long ago recognized the possible uses that this phenomenon might have in the fields of neurology, physiology, and psychiatry, only a few have actually availed themselves of the opportunities that it offers. The instruments that most workers used were difficult to operate and did not give reliable quantitative results; furthermore they gave either no graphic record at all, or a photographic record that required much time to develop and print. The shortcomings of these instruments may account at least in part for the lack of interest in this phenomenon. The instrument described below is free from most of these shortcomings, and furthermore it is readily constructed from standard equipment used in the electronics industry.

Fig. 1 shows a photograph of this instrument in operation. It consists of an amplifier (General Radio Company Type 715A direct current amplifier, operated on a 110 volt, 60 cycle supply), a 5 milliampere ink-recording ammeter (Esterline-Angus model AW), a control panel and a $4\frac{1}{2}$ volt battery. These parts are mounted on a stand equipped with roller-bearing casters. Fig. 2 shows a close-up view of the amplifier, recorder, and panel.

* Work carried out under a contract between the Office of the Surgeon General of the U. S. Army and the Johns Hopkins University.

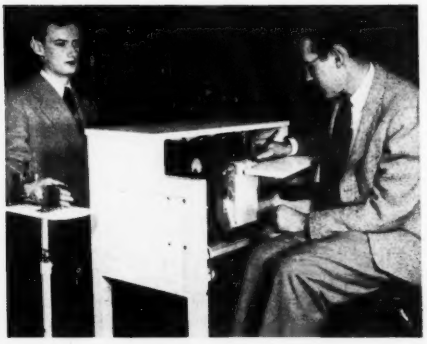

FIG. 1. Photograph showing skin galvanometer in use.

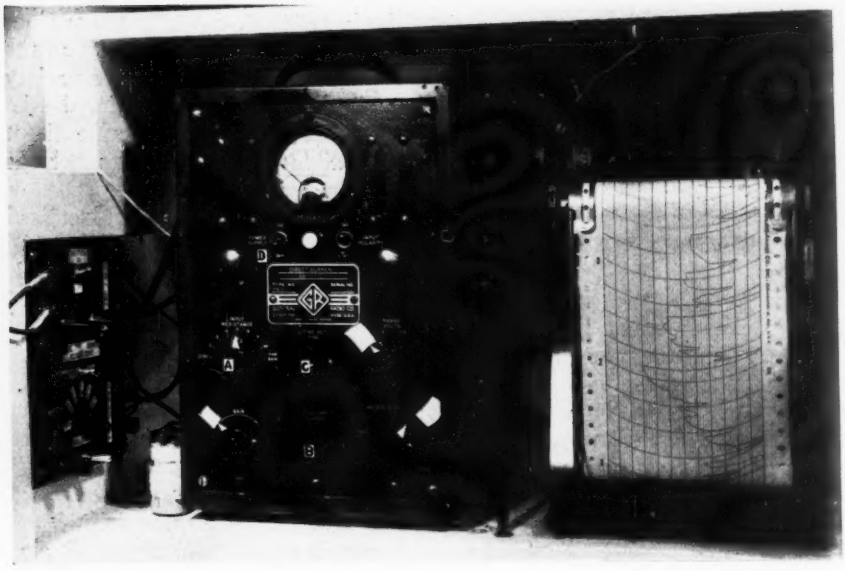

FIG. 2. Photograph showing amplifier, recorder, and panel.

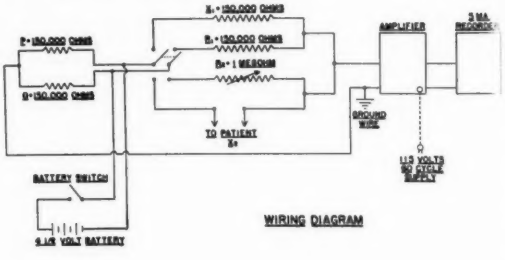

FIG. 3. Wiring diagram of skin galvanometer.

Fig. 3 gives the wiring plan. It is essentially a Wheatstone bridge circuit. The voltage supply comes from a $4\frac{1}{2}$ volt dry battery. The ratio arms P and Q consist of 150,000 ohm-$\frac{1}{2}$ watt carbon resistors, preferably a matched pair (obtainable at any radio supply store). This resistance level was chosen for the resistors since it closely approaches the average resistance of normal individuals and thus makes it possible to obtain a quick and accurate balance of the bridge. Two additional 150,000 ohm-$\frac{1}{2}$ watt resistors connected as the X and R arms make it possible to set the "zero" point on the detector-amplifier before the patient is connected into the circuit. A double-pole, double-throw toggle switch disconnects these 2 resistors and replaces them with the patient and 2 rheostats (1 megohm and 100,000 ohms), which serve to balance the bridge. The strength of the current that passes through the patient is not constant; it may vary between 2–20 microamperes, depending upon the resistance of the patient.

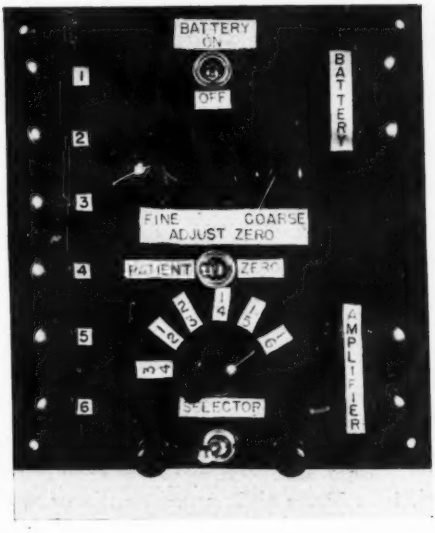

Fig. 4. Close-up view of panel.

The 4 fixed resistors; the coarse and the fine rheostats; the double-pole, double-throw switch; the single-pole, single-throw battery switch; an electrode selector switch; and 12 binding posts are mounted on the control panel (Fig. 4). Two of the binding posts make connection with the $4\frac{1}{2}$ volt battery and 2 with the amplifier. Six posts make connection with electrodes placed on 6 different parts of the body; a selector switch not shown on the wiring diagram connects various pairs of these electrodes to the measuring circuit. Two posts at the bottom center of the panel allow the subject to be connected with a dermometer which gives the skin resistance in ohms; a switch between these 2 posts disconnects the patient from the bridge circuit and establishes the connection with the dermometer. This dermometer is used during a test whenever it is desired to obtain an actual reading of skin resistance in ohms. The binding posts and switch are not essential to the operation of the skin galvanometer and are not shown on the wiring diagram.

The changes in the patient's skin resistance as they affect the currents in the bridge can be amplified to various degrees by the amplifier and recorded in ink on the recorder.

The paper moves at a speed of 3 inches per minute. Electrically operated signal markers at either side of the paper record the incidence of the various stimuli or other pertinent data.

An adjustable shelf (see Fig. 1) directly in front of the moving paper provides a hand rest and enables the examiner to write his notes directly on the record.

We have used 2 types of electrodes, either polished pure zinc discs $1\frac{1}{4}$ inches in diameter and a paste made of kaolin and saturated zinc sulphate solution, or the regular plates and electrode jelly that are used to record the electrocardiogram.

The following instructions give the steps involved in the operation of the galvanometer:

1. Attach electrodes to skin and connect to binding posts on control panel.

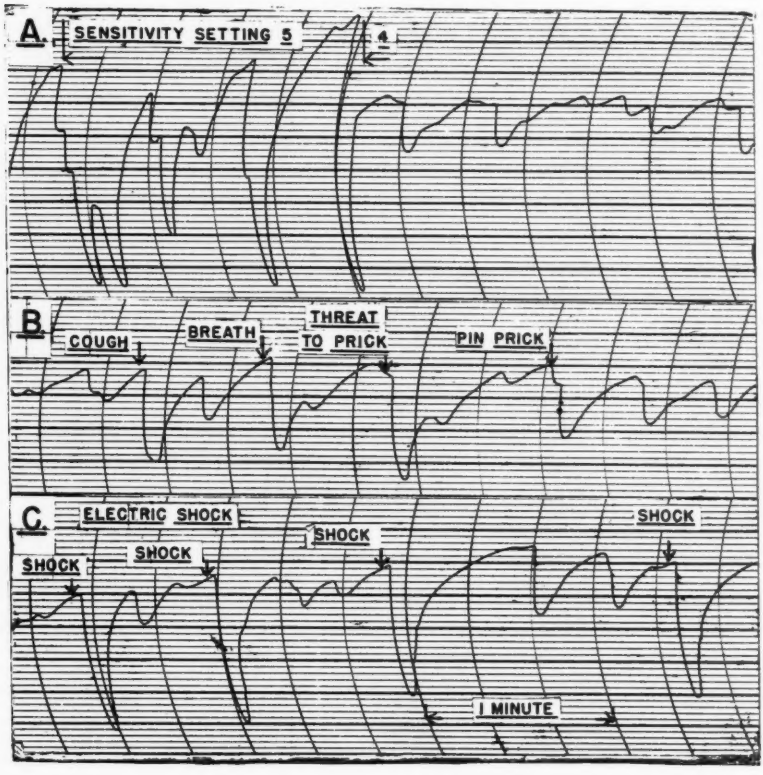

FIG. 5. Typical records showing spontaneous galvanic waves and responses to cough, deep breath, pin prick, and electric shock.

2. Turn on amplifier (Fig. 2—switch D) and allow to warm up for several minutes; turn on battery switch on panel.
3. Set double-pole, double-throw switch on panel to "zero."
4. Set sensitivity of amplifier at 4 or 5 (Fig. 2—knob A).
5. With "coarse-fine" zero adjusters (Fig. 2—B & C) on amplifier, set needle on recording meter to midscale position.
6. Throw double-pole, double-throw switch on panel to "patient" position. This connects patient and rheostats in X and R positions.

7. Adjust "coarse-fine" rheostats on panel until needle again rests on mid-scale position.
8. Start recorder and begin test.
9. If needle drifts off steadily to one side or the other, bring back with rheostat on panel until it stays constantly at midline position.
10. If needle fluctuations are greater than width of paper, reduce sensitivity until needle stays on chart.
11. Ask subject to cough or take a deep breath. These stimuli should give galvanic responses. If responses are too large and needle goes off paper, reduce sensitivity; if they are not large enough, increase sensitivity.

Fig. 5A shows several spontaneous galvanic fluctuations from a normal subject with electrodes attached to the palm and back of the hand. The cross lines give the time in 20 second intervals. The first half of the record shows the spontaneous waves with a sensitivity setting of "5"; the second half shows much smaller waves with a sensitivity setting of "4." Fig. 5B shows typical responses to cough, deep breath, threat to prick with pin, actual pin prick. Fig. 5C shows responses to electric shocks (induced current applied to leg).

We have used this instrument for the following purposes: (1) to test for the presence of sympathetic activity after sympathectomies; (2) before and after prefrontal lobotomies to compare the sympathetic responses to actual and threatened pain; (3) to test responses to direct stimulation of the sympathetic chain; (4) to test sympathetic activity in areas of referred pain; and (5) to record sympathetic responses that accompany pain.

In addition it has been used with a simple conditioning process to measure hearing ability in children and adults without the use of subjective reports;[1] it has also been used to record emotional responses elicited by standard and spontaneous questions in psychiatric interviews.

SUMMARY

(1) A simple and easily operated skin galvanometer constructed from standard parts used in the radio and electronics industries was described. It gives a graphic record of the activity in the sympathetic nervous system indirectly through the changes that sympathetic activity produces in the electrical resistance of the skin.

(2) This instrument has many applications in the field of neurology and neurosurgery as well as in psychiatry and other branches of medicine.

We acknowledge our thanks to Dr. William B. Kouwenhoven for his help and suggestions.

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CEREBRAL HEMISPHERECTOMY

REPORT OF A CASE TEN YEARS AFTER OPERATION

ERIC BELL, JR., M.D., AND LOUIS J. KARNOSH, M.D.
Cleveland Clinic, and the Frank E. Bunts Educational Institute, Cleveland, Ohio

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SINCE 1928 when Dandy described removal of the right cerebral hemisphere, this operation has not been enthusiastically received because of the generally discouraging results. In review of 13 cases of hemispherectomy in the literature, certain considerations have become clearer concerning (1) the selection of cases, (2) the technic of operation, (3) the resultant disability and ultimate prognosis of the patient, and (4) the proper evaluation of the procedure.

The indications for cerebral hemispherectomy are limited. It should not be considered except in cases of gliomas that are located well away from the midline and that have not invaded too deeply into the basal ganglia. This rules out the usual case of glioblastoma multiforme, with its propensity for advancing across the midline through the corpus callosum. Consequently, the indications narrow down to the occasional slowly growing oligodendroglioma and astrocytoma involving the middle portion of the hemisphere. If a tumor so situated is already causing progressive hemiparesis, the patient is more willing to accept the permanent postoperative disability.

Of the 5 patients operated upon by Dandy,[1,2] 1 died in 48 hours of hemorrhage; 1 died in 2 weeks from pneumonia; 1 died in 3 months with a recurrence, and 1 died in 3½ years with a recurrence. A follow-up study of the 5th patient beyond the 2nd postoperative week is not available.

Zollinger[17] described a patient who expired with meningitis 17 days after left hemispherectomy. Walker[15] reported a patient in whom the right cerebral hemisphere was removed by Evans, but the subsequent degree of disability was not detailed. Rowe[13] reported the case of a 38-year-old woman subjected to right hemispherectomy and emphasized particularly the psychiatric findings.

Dr. W. James Gardner, of the Department of Neurosurgery of the Cleveland Clinic, has resected the right cerebral hemisphere in 5 patients with brain tumor.[4,5,8,9,11] One died of meningitis 29 days after operation; 2 patients died with recurrence after 15 months and 3 years, respectively; 1 died 4 years after operation from an intracranial injury caused by a fall down cellar stairs. The 5th patient is still living 10 years after the hemispherectomy.

In contrast to the patients in Dandy's series, whose motor infirmities were severe, all 4 of our patients who survived the immediate postoperative period were able to walk unaided by cane or crutch and were able to dress themselves and to do light work after the operation. In all cases the relative

freedom from severe physical handicap and gross mental defect after removal of almost half the cerebrum was striking.

The outstanding clinical symptom in this artificial defect of the brain was a contralateral hemiplegia. This partial paralysis of the contralateral face and limbs, however, did not resemble in every way the classic spastic hemiplegia due to cerebrovascular thrombosis or hemorrhage. The chief difference was the manner in which the upper extremity hung loosely from the shoulder as the patient walked. The limb could be more easily manipulated than in the typical case of spastic hemiplegia. However, on the affected side the tendon reflexes were hyperactive, and the Hoffmann response was consistently positive in all the patients. All patients could elevate the shoulder and had some ability to abduct and adduct the arm and to flex and extend the forearm at the elbow. However, there was a uniform lack of power of the wrist movements and of the fingers. A weak grasp was the maximum response of the affected hand even years after operation. The opponens function of the thumb was entirely absent, and voluntary movement of the individual fingers was impossible.

On the lower left extremity these patients demonstrated a fairly well-retained voluntary control of both thigh and leg movements. Volitional control of the ankle and toes was absent. In every instance on the affected side the tendon reflexes were hyperactive, ankle and patellar clonus could be elicited and there was a strongly positive Babinski toe reflex.

With all these abnormal signs on the upper and lower limb, evidence of impairment of axial muscles of the neck, spine, chest, and abdomen was lacking. For instance, none of the patients demonstrated marked disparities in movement of girdle muscles, in the action of the ribs during respiration, or in the tone of the muscles of the abdominal wall.

The degree of facial palsy in these patients showed considerable variation. In some instances the left angle of the mouth was immobilized to both voluntary and mimetic stimulation. In other cases the facial weakness was almost imperceptible. In no patient did the facial paralysis involve the upper portions of the face as it does in peripheral or Bell's palsy.

This unique type of hemiplegia is the result of a mechanically induced brain defect of known extent. Decortication of one side of the body produces the greatest disability in the mouth, the hand, and the foot. These parts have the most completely lateralized representation in the cortex. Hughlings Jackson[3] expressed this conviction in his early observations of cortical function. He stated that movements of the fingers, which have a most specialized and highly differentiated motor behavior, have the least bilateral cortical control.

Sensory disturbances in these patients were not as profound as one might conclude from textbook concepts. It was evident that the type of sensation which was preserved was that which has been best described by Head[7] under the term "protopathic or vital sensation." The more profound loss consistently occurred in that type of sensation which he designated as "epicrit-

or gnostic sense." This loss of epicritic sense, however, did not apply to the trigeminal area of the affected side, for in all cases two-point discrimination and awareness of light touch was preserved over the skin of the face, although this type of sensitivity was never as good as it was on the unimpaired side.

This preservation of so-called cortical or epicritic sense over the trigeminal zones can best be explained on the basis of bihemispheric representation. Experiments by Woolsey and Fairman[16] indicate that even in certain mammals there is evidence of bicortical representation of trigeminal epicritic sensation. They postulate two sensory areas in each hemisphere. Their "sensory area I" receives impulses from both trigeminal areas as well as from the entire contralateral half of the body. However, even in the decorticated half of the body below the trigeminal zones of our patients there was some evidence of a preservation of sensory functions which are ordinarily assigned to the cortical levels. The patients were able to localize the site of a painful stimulus within 2 or 3 metameres. They were also able to recognize roughly in what position the decorticated arm or leg was passively placed. Vibratory sense was preserved over the entire affected side, although it was moderately impaired. These evidences of some preservation of position sense, of vibratory sense and of definite although poor localization of painful stimuli seem to indicate that there may be, as Woolsey and Fairman have suspected, some ipsolateral corticalization of the old sensory system in their "sensory area II" which, they believed, received impulses from both sides of the body.

Probably of greater concern to the neurosurgeon is the danger of global aphasia when there is uncertainty as to which hemisphere controls speech function. Zollinger[17] is apparently the only writer who recorded the results of a left hemispherectomy in a right-handed patient. In this case the aphasia was incomplete. Several hours after operation the patient answered "all right" to all questions and showed her teeth on command. The following day she became more alert than before operation and added "yes" and "no" to her vocabulary. On the 2nd postoperative day "thank you" and "sleep" were added. On the 3rd postoperative day she used the words "good-bye" and "please" and showed a more accurate use of words. The next day she used "yes" and "no" apparently with a full understanding of their meaning. Zollinger says, "It was my impression that vocabulary could have been gradually increased by training." Evidently an elementary vocabulary was retained in the non-dominant hemisphere and was increased somewhat by postoperative guidance. Nielsen[10] and others have collected evidence indicating that Broca's area is not invariably located in the "dominant" hemisphere and that apparently there are some people in whom the function of Broca's area has shifted from one hemisphere to the other. On record are several cases in which "handedness" was located in one cerebral hemisphere and "speechedness" in the other. Apparently such a phenomenon occurred in the patient to be presented.

Two of our patients who were tested with an audiograph after hemispherectomy showed an almost uniform loss of 5 to 10 decibels in both air and bone conduction in the contralateral ear. This is in agreement with Tunturi's[14] findings on audio-frequency localization in dogs. He concluded that each ear is represented bilaterally in the auditory cortex of each hemisphere but that this representation in each cortical area is stronger for the contralateral ear.

All patients studied by visual field examination showed a homonymous hemianopsia after cerebral hemispherectomy. Dandy[2] indicated that the hemianopsia was complete in the 3 patients whom he studied. All 3 of our patients in whom visual fields were charted showed definite sparing of macular vision. Rowe[13] also recorded macular sparing in his case. Reports by Penfield and Evans[12] indicate that, with fair uniformity, the macula is spared bilaterally when only one occipital lobe is removed. When the optic tracts are unilaterally interrupted anterior to the occipital lobes there may be either macular sparing or macular splitting. Apparently the result depends entirely on the individual affected. It seems reasonable to assume that the macular fibers may be more completely represented bilaterally in some persons than in others.

Mental changes following removal of most of one-half of the cerebrum are surprisingly mild. Rowe[13] described in detail the mental deviations of a female patient. For several weeks after operation she lacked normal inhibitions. There was a wide range of mood and a temporary impairment of recent memory. She gradually became more normal over a period of 6 months until she showed only a slight lability of mood and a talkative disposition. She was mentally active and her intelligence quotient was the same as it had been 1 year previously. Dandy[2] stated that his patients showed no evidence of impairment of intelligence or of memory, although some were more facetious than they had been prior to their illness and had less ability to concentrate for a long period of time. Penfield[12] stated that the defect produced by a frontal lobectomy is "a lack of capacity for planned administration" and "the loss of power of initiative." Study of our cases has indicated that this is the most accurate description of the mental defect produced by removal of the right cerebral hemisphere. None developed antisocial tendencies or became a public charge.

The major complication in many of the recorded cases of cerebral hemispherectomy was the development of postoperative meningitis, which caused the demise of the patient. It should be emphasized that these operations were performed prior to the time that penicillin, streptomycin, and many of the sulfonamide agents became available.

CASE REPORT

A white man, aged 35, was admitted to the Cleveland Clinic in January, 1938, because of a history of progressive weakness of the left leg for the previous 2 months, complicated by jacksonian motor seizures starting on the toes of the left foot. There was some weakness of the left arm, and tendon reflexes were hyperactive on the

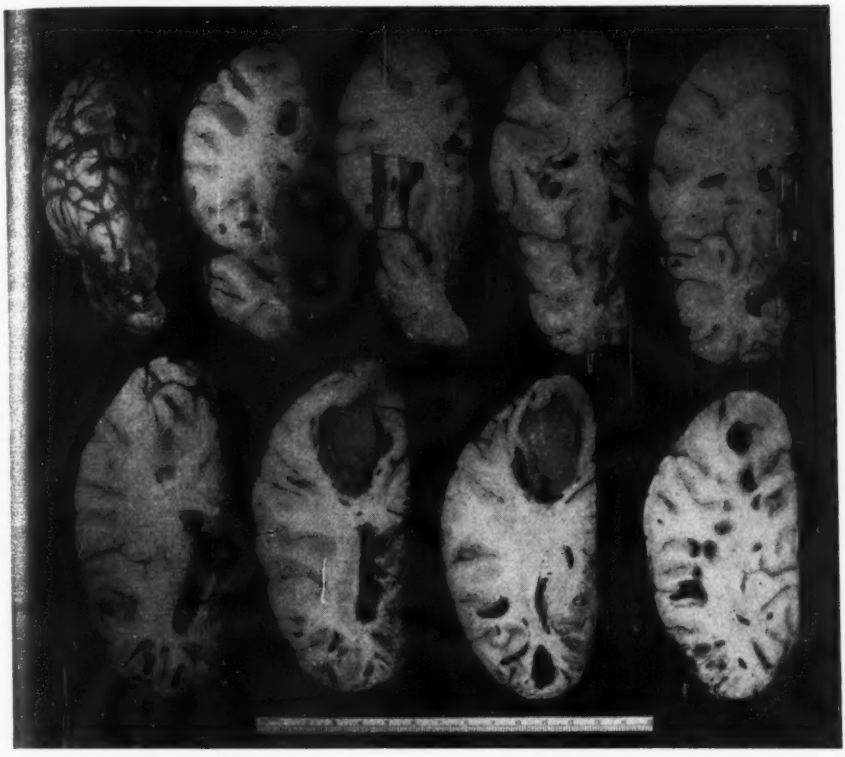

FIG. 1. Coronal sections of the right hemisphere which was removed from the patient 10 years ago.

The astrocytoma occupies the white matter in the 3rd frontal convolution, anteriorly, and extends upward and posteriorly into the white matter of the parietal lobe, where it assumes its greatest calibre and where there is evidence of cyst formation.

The line of surgical section shows that the caudate nucleus, the putamen and even the outer portion of the globus pallidus have been sacrificed in carrying out the hemispherectomy. No tumor tissue is found along the plane of surgical incision, the dark and mottled areas being due to hemorrhage incidental to operation.

left. There was no evidence of a left facial weakness. He had no choking of the disks and no sensory disturbances. Visual fields were normal. A right frontoparietal craniotomy revealed the presence of a subcortical astrocytoma. There was doubt as to the advisability of removing the right cerebral hemisphere because the patient was definitely left-handed. He sighted with the left eye and wrote with the left hand. He used his right hand very clumsily and without evidence of ambidexterity. His father also was left-handed. It was assumed that the patient's motor speech center was located on the right side. The problem was solved by trephining under local anesthesia over the supposed area of Broca in the right hemisphere and by injecting novocaine subcortically into that area until the left angle of the mouth was paralyzed.[6] Fortunately, the patient's speech was unaffected by this procedure, and it was there-

fore concluded that the patient had an intact motor speech center in the le cerebral hemisphere while his dominant hand center was in the right hem sphere.

On Mar. 3, 1938, most of the right cerebral hemisphere was removed. The re sected specimen contained the claus trum, the putamen, most of the cau date nucleus, and approximately th outer 1/5 of the globus pallidus (Fig. 1). The chief structures remaining *in situ* on the right side of the brain were the hypothalamus, thalamus, and about 4/5 of the globus pallidus. Mesial portions of the hippocampal and fusiform gyri were spared also. The postoperative course was essentially uneventful. Three weeks after operation the patient developed sufficient use of the left leg to enable him to walk with some assistance. There was a complete paralysis of his left arm and a marked weakness of the left side of the face, most noticeable about the mouth. The mental symptoms in the immediate postoperative period were simply those of emotional blunting and some mental retardation. Speech was not affected.

FIG. 2. Left hemiplegia in a patient with a right cerebral hemispherectomy of 10 years' duration. The subject is demonstrating the maximum degree of voluntary elevation of the left arm as compared with the right. When at rest the left arm and hand hang loosely from the shoulder. The facial expression demonstrates an animia of the left side. The left leg is swollen by varicosities.

The patient returned home and began a long period of readjustment. He undertook his old job of operating a small restaurant with the assistance of his wife. He was noted to be jocular, facetious, and fond of teasing his friends. His wife insisted that this was his normal behavior. He had some difficulty in concentrating and was not greatly concerned about the financial affairs of the family. He was inclined to dispose of serious matters with a shrug of his shoulders or with a nonsensical remark. Because of poor control over his right hand he was almost helpless in dressing himself, eating, and writing.

After several months he taught himself to walk without the aid of a cane. Gradually there became possible more individualized movements, such as crossing the left knee over the right, elevating the left shoulder, and even grasping feebly with the left hand. By great perseverance he has finally developed the dexterity of the right hand to the point where he is able to dress himself, to button his clothes, to use eating utensils, and to write his name. He has become rath

proficient in the use of his right hand in chopping wood and in doing odd jobs about the yard. Whereas he formerly was helpless when he fell to the ground, he now has developed a technic of picking himself up entirely unassisted. This notable improvement of motor function has been the result of a gradual readjustment over a 10-year period. Demonstrable improvement is continuing at the present time (Fig. 2). He would like to drive a car but is deterred from this by the combination of hemianopsia and hemiparesis.

EEG studies show a marked decrease in amplitude of the wave pattern over the right side of the head and a total absence of normal alpha activity from that region. Tracings from the left cerebral cortex show normal rhythm.

The sensory examination of this patient 10 years after hemispherectomy is informative. Sensation over the entire right side is normal and is intact to a line 2 cc. to the left of the midline. He complains of a "heavy, dull, doughy, numb feeling" over his left side from the neck down. He states that the left side of his face does not feel exactly like the right side, and explains, "It takes longer for a feeling to come through on the left side of my face." He has no spontaneous pains. The light touch of cotton wool can be perceived on the face but not on the remainder of the left side. When a camel's-hair brush is dragged along the skin on the left side of the body it is appreciated as a collection of "unpleasant small stickers or pins." With his eyes closed, the patient states that this moving brush is experienced as a stationary or twisting stimulus.

Localization was tested by having the patient place the forefinger of his right hand on the area of his skin stimulated. Light touch and pin prick were accurately localized in the area supplied by the left trigeminal nerve. In testing the remainder of the left side it was necessary to use a painful stimulus, because light touch was not appreciated. The patient complained bitterly throughout this testing period because he thought that the examiner was pressing harder than was necessary. A firm pressure with a dull object caused the identical type of pain that was elicited by a pin prick. The patient was able to localize these noxious stimuli remarkably well in the upper left portion of the body, especially in the distal portion of the arm, where he consistently designated the correct area within 5 cm. On the upper part of the body and the proximal portion of the arm he usually was correct within 15 cm. This ability to localize stimuli on the left side became progressively poorer toward the lower portion of the body and leg.

It was our impression that when a particular area on the left side was stimulated the patient was able to localize the stimulus to its particular metamere more accurately than to the designated area within the metamere. For instance, a stimulus to the lateral surface of the left knee was occasionally referred to the dorsum of the left foot but never to the medial surface of the same knee. The patient stated that stimuli on the left side below the face area were localized by the prolonged after-pain which lingered many seconds after the stimulus had been withdrawn.

Tickling and scraping lightly with the fingernails over the patient's left palm and sole caused a very unpleasant burning, stinging feeling. Vibration sense over bony prominences was definitely preserved, although it was slightly diminished in the left leg and arm as compared with the right.

The patient showed a marked loss of two-point discrimination. This loss seemed to be entirely out of proportion to the loss of localizing ability for one point. When two points 5 to 10 cm. apart were employed, he could tell occasionally whether one point or two points were used. He recognized this, he said, only by the fact that "Two points hurt twice as much as one point." He was not able to localize two points

applied simultaneously to the left side below the face unless they were separated by several dermatomes.

The patient was not able to distinguish cool water from tepid water consistently, but either hot or cold water was quickly perceived as a painful stimulus. He has noticed that for these extremes of temperature there are often no differences in sensation which allow him to distinguish hot from cold; they are both painful. He is unable to enjoy winter outings because the cold causes his left side to become disagreeably painful, and when he becomes chilled there is a severe involuntary, persistent clenching of the left hand and flexion of the left arm at the elbow.

Passive movement was appreciated only if there was a sudden motion in a definite direction over a range of at least 15°. The arm or leg frequently could be moved over a distance of 20 to 25° without the patient's knowledge if the change was made slowly and easily. If the movement was appreciated, the direction was consistently stated correctly. When the examiner placed the left limbs in a certain position, the patient was able to simulate this position fairly well with the limbs of the unaffected side. Position sense of the fingers and toes was entirely lacking. The patient stated that sensibility to pain was the same in both testes and on both sides of the glans penis. Abdominal and cremasteric reflexes were present bilaterally but were diminished on the left side. Strongly positive Hoffmann and Babinski responses were obtained on the left. Wrist, patellar, and ankle clonus could be elicited in the left extremities. The reflexes on the right side of the body were normal. Bladder and bowel function, sphincter control, and sexual activity were unaffected by operation.

Procaine hydrochloride block of the right stellate ganglion was performed. A good physiologic response was indicated by the resultant Horner's syndrome on the same side. The patient spontaneously stated that the left side of his body had "more feeling in it and seemed 80 pounds lighter" following the block. He later claimed that the left arm and leg felt "easier to handle" for several weeks after the block. Objective confirmatory evidence of improvement could not be demonstrated. While no exact conclusions can be made from this experiment, it is surmised that the blocking of the sympathetic impulses to the right side of the head affected the circulation of the intact right thalamus sufficiently to cause the modification of sensation from the paralyzed side as the patient described it.

A conference with the wife of the patient gave us the clearest picture of his mental status 10 years after operation. He still worries little about finances and may spend his extra money with rather poor judgment at times, but he has never shown any asocial or antisocial tendencies. He takes home life seriously and continually occupies himself about the house and yard. He is personally immaculate, as he always was, and continues to keep his room in order and to store his belongings in their proper places. He likes to make short journeys, and he occasionally rides a truck to neighboring towns with his friends. He occasionally accompanies his cronies on trips to a hunting lodge where he stays in camp and tends the fire. He continues to be sociable and to amuse everyone with numerous stories. His disposition is generally good. He becomes emotionally upset easily, but these moods pass quickly. He has never exhibited a hypochondriacal attitude. His memory does not seem to be impaired. His wife states that he is still a good poker and pinochle player. He does not use intoxicating beverages to excess and does not show a decreased tolerance to alcohol. His health has been excellent. Both he and his wife are grateful that the operation was performed.

CONCLUSION

In carefully selected cases of slowly growing gliomas, cerebral hemispherectomy may be the treatment of choice. The sensory and motor postoperative changes are not utterly incapacitating if the paleostriatum and thalamus remain essentially intact. Mental changes are not profound.

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TRAUMATIC SUBDURAL HEMATOMA—ACUTE, SUBACUTE AND CHRONIC

AN ANALYSIS OF SEVENTY OPERATED CASES

FRANCIS ECHLIN, M.D.*
New York, N. Y.

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In the past it has been common practice to class all subdural hematomas as acute or chronic. Acuteness has been estimated on the basis of elapsed time from the injury, or on the degree of associated brain damage. According to one classification, all hematomas that cause death or come to operation before the 21st day after a cranio-cerebral injury are described as acute (Kennedy and Wortis[9]). In another classification, cases of subdural hematoma that still show unhealed or acute brain injury are placed in the acute group, the others being classed as chronic (Munro[15]). In still another classification, an acute subdural hematoma is regarded as the result of a severe head injury, usually with a fracture of the skull and more or less extensive brain laceration, whereas a chronic subdural hematoma is considered to be unassociated with severe brain injury and the history of trauma, as a rule, is slight or absent (Peet[17]).

As Munro[15] has pointed out, any classification of subdural hematomas into groups is artificial, for as Munro,[13] Laudig, Browder and Watson,[11] and King[10] have clearly demonstrated, the chronic form of traumatic subdural hematoma, so well described in the literature[3,6,8,12,14,18,19,20] is nothing more than a later variant of the acute phase.

Acute hematomas become chronic only gradually, and progressive symptoms appear as the hematoma enlarges by the acquisition of further fluid, usually through the process of osmosis.[5,16,21] Therefore if any classification is to be used it should clearly indicate that there is a gradual transition between the so-called acute and chronic hematomas.

The purpose of this paper is to combat the present tendency to class all hematomas as acute or chronic. Such a classification is misleading and fails to emphasize the existence of a large group of subdural hematomas which, neither on clinical nor pathological grounds, can be satisfactorily classed as acute or chronic. If this is not clearly recognized, the average observer may fail to diagnose the subdural hematoma that ends in a fatality about the end of the first week to several weeks after injury.

In reviewing the cases of subdural hematoma to be presented, it is apparent that they fall roughly into three classes, on the basis of the clinical course, type of hematoma found at operation, and degree of associated brain injury; namely, acute, more or less subacute, and chronic. There is con-

* 555 Park Avenue, New York 21, N. Y.

siderable overlap between these three classes of hematoma, on the basis of the criteria mentioned, but the existence of a subacute variety (as first suggested by Bucy,[2] Gurdjian and Webster[7]) as well as an acute and chronic one, is borne out on analysis of the findings in patients operated upon at different time intervals following trauma.

MATERIAL

The material presented is based on a study of 70 patients with subdural hematomas operated upon during the 20-month period from January, 1941 to July, 1942, on the Neurosurgical Service, 3rd Surgical (New York University) Division at Bellevue Psychiatric Hospital. During this interval approximately 7916 patients entered the hospital with a history or signs of recent head injury. Of these, 1415 showed signs of intracranial injury, considered severe enough to admit them to the neurosurgical ward. It was among this group that most of the 70 subdural hematomas were found. A few of the more chronic ones, however, were transferred from the psychiatric or medical services, having been received there with a diagnosis other than head injury.

These statistics show that the incidence of subdural hematomas was less than 1 per cent in the entire series of head injuries, and less than 5 per cent in the group of cases of severe head trauma.

Of these 70 patients with subdural hematoma, 50 were operated on by me, and 20 by Henry Wigderson, but with 3 exceptions I followed them all pre- and postoperatively.

Thirty-five additional patients, suspected of having a subdural collection of blood as the cause of their symptoms, were explored. In many of these cases blood was found in the subdural space but they were not classified as subdural hematomas.

In order to emphasize the difference in the clinical course of the illness, and in the pathological nature of the hematomas in the various patients, they will be analyzed in groups according to the time elapsed since injury. Analysis of other data pertinent to the diagnosis and treatment of subdural hematoma will follow.

The clinical course and nature of the hematoma found in patients operated upon within 24 hours following trauma

There were 10 patients in this group. Trauma was severe in all and consciousness was lost immediately following injury. At no time did a lucid interval follow. The condition changed from coma to stupor in 5 patients, and to semi-stupor in the remaining 5. After thus showing some degree of improvement, stupor became progressively deeper in each instance. The hematoma removed at operation consisted of a large, currant-jelly-like clot with some fluid blood in 9 cases, and fluid blood alone in 1. Of the 10 patients in this group only 1 survived. The others all died within 5 days. Autopsy revealed extensive traumatic brain injury in all cases.

Comment. These 10 patients suffered from a severe brain injury plus a fresh subdural hematoma. Death in 9 cases was due almost if not entirely to direct brain trauma, the hematoma, although large, being incidental. Survival in 1 case may be attributed to removal of the hematoma.

The clinical course and pathological nature of the hematoma in patients operated upon 2 to 7 days after injury

There were 11 patients with subdural hematoma operated on 2 to 7 days after injury. Trauma was severe with immediate loss of consciousness in 9 cases. Two patients had moderate trauma and did not lose consciousness.

Nine of the patients were admitted to the hospital immediately after injury, 1 was brought to the hospital 12 hours following injury, and 1 in 24 hours. All the patients remained on the neurosurgical ward until operation.

One of the patients continued to be comatose from the time of injury to operation on the 3rd day. Eight patients, after recovering consciousness, were very drowsy and disoriented and subsequently became much more drowsy or stuporous prior to operation. In 5 cases the onset of sustained stupor was sudden and in 4 of these the stupor was precipitated by a seizure. One patient had a lucid interval for 5 minutes after injury and then rather rapidly became stuporous. Stupor persisted 24 hours and was replaced by drowsiness which progressively deepened. The final patient did not lose consciousness with injury but went home in an alcoholic condition; 19 hours later he had a seizure, became comatose for about 1 hour and following this was confused, drowsy and aphasic; 24 hours later he had another seizure, became progressively comatose and was operated upon.

The hematomas removed at operation from the 11 patients consisted of relatively fresh blood, in the form of large (9 cases) or small (2 cases) currant-jelly-like clots, accompanied by varying amounts of dark brown to black fluid.

Four of these patients survived and 7 died.

Comment. The clinical course, as well as the pathological nature of the hematomas, was acute or relatively acute in these 11 patients. The clinical course in most cases was directly influenced, in an important manner, by an associated brain injury.

The clinical course and nature of the hematoma in patients operated upon from the 7th to the 21st day following injury

There were 30 patients in whom a large subdural hematoma was found at operation 7 to 21 days after injury. In 19 cases operation was performed on the 10th to the 15th day following trauma.

A history of head trauma was obtained in all cases. In 27 the trauma was accompanied by immediate loss of consciousness. In 2 the injury was mild without loss of consciousness, and in 1 the history is indefinite. Twenty-four of the patients were brought to the hospital within a few hours of the

accident. The other 6 were admitted within 8 days following injury. All of the patients, with 1 exception, were under observation on the neurosurgical ward throughout the course of their illness. The exception was a patient who signed out of the hospital on the 5th day after injury, to be returned in stupor 8 days later.

One of the most important clinical signs in arriving at a diagnosis in this group of 30 cases was the state of consciousness. All of the patients who lost consciousness after injury subsequently regained it. Ten of the patients remained drowsy and disoriented throughout, and before operation was undertaken drowsiness had become progressively deeper.

Fourteen patients became mentally alert and cooperative but showed some degree of disorientation. Each of these 14 individuals later went through a period of progressive, usually fluctuating drowsiness preceding operation.

Only 6 patients could be described as reaching a state of mental clarity following their injury, but 4 of these subsequently became drowsy and then suddenly stuporous prior to operation. In 3 instances the stupor was precipitated by a convulsive seizure. In the remaining 2 patients the diagnosis was made by encephalogram before the onset of severe symptoms.

On a pathological basis it was found at operation that 15 of the hematomas, in this group of 30 patients, more closely resembled the so-called "chronic" than the "acute" variety of subdural hematoma, and consisted of dark, coffee-colored fluid containing, in 5 cases, small, soft blood clots in various stages of liquefaction. In 6 of these 15 patients thin membranes surrounded the hematoma, in 1 case as early as the 8th day after injury. The other 15 hematomas varied in consistency from currant-jelly-like clots with black, tarry or brown fluid to 2 cases of so-called subdural hydroma containing canary-yellow fluid.

In this group of 30 patients, 23 lived and 7 died.

Comment. This entire group of 30 patients, operated on 7 to 21 days after an acute injury of the brain, would ordinarily be classed as having an acute subdural hematoma. However, 20 of the patients followed a subacute clinical course, in that they became alert, cooperative and showed signs of improvement for a week or more. In 15 of the 30 cases the pathological nature of the hematoma removed at operation more closely resembled that seen in "chronic" cases.

Progressive symptoms and signs of brain dysfunction in most of the cases was due to a subdural hematoma. Trauma may have disturbed the normal physiology of the brain and rendered it more susceptible to the pressure from a hematoma in some instances, but, in the majority, traumatic brain injury itself was not the cause of progressive neurological symptoms and signs. On clinical and pathological grounds then, as well as on the degree or part played by an associated brain injury, many of these cases are best described as subacute.

The clinical course and nature of the hematoma in patients operated on from the 22nd to the 31st day after injury

There were 10 patients in this group and a history of trauma to the head was obtained in all. Nine patients lost consciousness with injury and in 1 there was no history of unconsciousness. Five of the patients were taken directly to the hospital from the scene of the accident and remained until after operation. Three were admitted 3 to 20 days after injury. Two were admitted immediately following injury, left the hospital in several days, and were readmitted 25 to 27 days later.

Of the 10 patients, 4 became mentally clear for varying periods and the remainder became alert but showed some memory defect or disorientation. Five of the patients later showed a progression in their disorientation with drowsiness. Five did not show progressive drowsiness, but other features in their clinical pictures suggested the possibility of a subdural hematoma and the diagnosis was confirmed by encephalogram.

At operation the hematomas in the 10 patients resembled the usual fluid ones seen in many chronic cases, but 4 patients had in addition soft, brownish-colored clots and 1 patient, operated on 31 days after injury, an organized hematoma of the consistency of liver. Membranes were present about the hematoma in at least 4 cases.

All of the 10 patients in this group survived.

Comment. According to one classification, all of these patients would be regarded as suffering from a chronic subdural hematoma since they were operated on more than 21 days from the time of injury. According to another classification, many of the patients would fall into the class of acute subdural hematoma, since the majority of them had a relatively recent brain injury which certainly was not completely healed in all cases.

Actually, at least 5 of the 10 patients followed a subacute clinical course and, although they became alert for varying intervals, nevertheless remained disoriented and later showed progressive disorientation and drowsiness before operation. Five other patients in this group, in whom a suspected hematoma was verified by encephalogram, could be classed as either subacute or chronic, but certainly not as acute on the basis of the clinical course nor pathological findings.

CHRONIC SUBDURAL HEMATOMA

Nine patients in the series were classed as suffering from a chronic subdural hematoma. A history of severe trauma was obtained in only 2 cases and these individuals were admitted 30 to 42 days thereafter. No definite history of trauma was obtained in the remaining 7 patients, although this does not exclude such an etiology.

Eight of the patients had shown progressive drowsiness before they were seen by the neurosurgical staff. Six of them were disoriented, 1 appeared psychotic and 2 were in coma on admission to the neurosurgical ward. Only 1 patient was alert and cooperative pre-operatively. The diagnosis of an in

tracranial lesion in this patient and 1 other was made by encephalogram.

All of the 9 patients had hematomas which were fluid with surrounding membranes. In 7 cases the fluid was of a brown color and in 2 it was yellow. A few mm. of soft, shaggy, brown clot was present on the inner surface of the outer membrane in 4 cases.

Seven of the patients survived and the 2 who were in coma at the time of operation died.

LOCALIZING SIGNS IN SUBDURAL HEMATOMAS

All of the 70 patients described in this paper showed abnormalities in the neurological examination.

In 29 cases there were localizing signs which correctly indicated the side of the hematoma, but in 12 the signs pointed to a cerebral lesion on the side opposite the hematoma. In 21 cases the neurological findings were inconclusive so far as localization was concerned. There were 8 cases of bilateral subdural hematoma. In 3 of these the neurological examination indicated a probable unilateral lesion, and in 5 the signs were not localizing.

The most common signs present were enlargement of a pupil, a slight facial weakness of central type, a diminished abdominal reflex unilaterally, inequality in the deep reflexes, a positive Babinski, and a slight weakness of one arm or leg. These signs when present from the time of admission were of little value in diagnosis or in localization. Signs that appeared at a later date, however, and especially when progressive, were of distinct value in making the presumptive diagnosis of subdural hematoma.

Marked hemiparesis to hemiplegia was present in 14 cases. In 8 patients the weakness was on the same side as the hematoma and in 6 on the opposite side. In no case was there positive evidence that a marked hemiparesis or hemiplegia was due solely to a subdural hematoma, with the exception of 2 cases of chronic hematoma. This is a most important point in the differential diagnosis between subdural hematoma and spontaneous or traumatic intracerebral lesions.

In 9 cases a marked weakness of one arm or leg was present from the time of admission, shortly after injury, and direct brain trauma probably was the chief causative factor. Of these 9 cases, 5 were among the patients operated upon in the first 24 hours. In 3 patients, who had moderate weakness of one side, a hemiplegia developed following a convulsive seizure.

Localizing value of a unilateral dilatation of the pupil. Inequality of the pupils was noted in 48 patients with a unilateral subdural hematoma. In 24 patients, at the time of operation, the larger pupil was on the same side as the hematoma and in 24 on the opposite side. In many of these cases there had been some fluctuation in the relative size of the pupils.

A relatively marked dilatation of one pupil was present at the time of operation in 25 cases. In these patients the large pupil was on the side of the hematoma in 14, and on the opposite side in only 7 cases. Quite marked enlargement of one pupil, however, was also noted in 4 patients with a bilateral hematoma.

From the above it is seen that a marked dilatation of one pupil is of some localizing significance, whereas a slightly dilated pupil is of no value in indicating the side of the hematoma.

CONVULSIVE SEIZURES

Convulsive seizures occurred in 17 patients. In the patients under discussion it is impossible to say whether the seizures were due to direct brain injury, alcoholism (of which there was a history in the majority of patients) or the subdural hematoma. Some of the patients may have had chronic epilepsy. However, the high incidence of convulsive seizures is a serious complicating factor in the presence of a brain injury and subdural hematoma, and frequently precipitated a hemiparesis or a continuing stupor in an already drowsy patient.

SUDDEN ONSET OF STUPOR OR COMA

In 11 patients, who were already drowsy, there was a sudden onset of persisting stupor or coma, and in 7 this was preceded by a convulsive seizure. Four of the patients in whom a seizure precipitated stupor were among the group operated upon on the 2nd to the 6th day after injury.

THE OPTIC FUNDI

Papilloedema was uncommon in this series of cases during the first 2 weeks after head trauma.

CAUSES OF DEATH

There were 25 deaths among the 70 operated cases. Of these, 9 occurred in the group operated upon in 24 hours after trauma, and may be attributed largely to associated brain injury.

Of the remaining 16 patients, 10 were in coma and 4 in stupor at the time of operation and were helped little by the removal of the hematoma. Five of the patients operated upon before the 31st day after trauma could probably have been saved by earlier operation, and this is certainly true of the 2 with chronic subdural hematoma.

It is very evident that if a patient is allowed to become comatose or deeply stuporous his chances of survival are greatly lessened even though the hematoma is evacuated.

Evans and Scheinker's[4] demonstration that an expanding lesion may produce actual thromboses of large vessels as a result of herniation of cerebral tissue over the free edge of the tentorium or beneath the falx, may explain the coma and lack of recovery following operation in these cases.

At the time of operation 11 patients were comatose (exclusive of those operated upon in the first 24 hours) and 10 of them died; 13 patients were stuporous and 4 of these died.

One patient died of an operative infection and 1 of meningitis 4 months postoperatively.

PULSE, CEREBROSPINAL FLUID PRESSURE AND BLOOD PRESSURE

During the study a quite detailed analysis of the relationship between blood pressure, pulse, cerebrospinal fluid pressure and state of consciousness has been made, but will not be presented in detail here. The findings are in keeping with those reported by Browder and Meyers.[1] In summary: a pulse rate below 60 was a common finding in subdural hematoma, being recorded in 32 cases, and was noted in the presence of a normal CSF pressure in 6 cases. In 13 cases when the CSF pressure was over 300 the pulse rate was over 60.

The CSF pressure was recorded on the day of operation in 48 cases. In 15 patients the lumbar puncture pressure was below 160 and in 33 above this level. In 17 patients the pressure was above 290 and in 13 it was 300 mm. of water or over. The lumbar CSF pressure bore no direct relationship to the state of consciousness. It was normal or below normal in 10 patients who were stuporous and elevated in many patients who were alert. The blood pressure likewise showed little if any relationship to the CSF pressure except perhaps in the first 24 hours after injury.

INCOMPLETE REMOVAL OF SUBDURAL HEMATOMA

In some cases, so-called recurrent subdural hematomas are due to incomplete removal of the clot at the first operation. The following cases are examples of incomplete surgical removal of a hematoma.

Four patients were operated upon on the 7th to the 14th day after injury. In each case the subdural space was exposed through bilateral posterior parietal burr holes and a subdural hematoma was found, consisting of coffee-colored fluid and a few clots. A thin membrane was present in 3 of the cases. In all patients a large quantity of the dark coffee-colored fluid was obtained, and it was felt that the patient's hematoma had been adequately removed. In addition a rubber tissue drain was left in the subdural space, through a stab wound in the scalp, for 24 hours.

Two of the patients improved temporarily, but within 1 to 2 weeks became progressively drowsy and presented other signs suggesting the presence of subdural hematoma. The other 2 patients failed to improve following operation. These 4 patients were all re-explored through a subtemporal decompression on the side of their original hematoma. In each case a large currant-jelly, or milk-chocolate type of clot, plus old fluid, was found.

Three of the patients survived and 1 died.

It was this type of experience that indicated that some subdural hematomas, although they may appear to be largely fluid, may contain large clots which cannot be adequately removed nor visualized through the posterior parietal approach.

OPERATIVE APPROACH

Following the above-mentioned experience it became our custom to explore for all subdural hematomas through the bilateral temporal approach. With the patient in a semi-sitting position supported by a small posterior

neck rest, a burr hole is made just anterior to and above each ear. The patient, however, is draped so that subtemporal decompression and occipital burr holes for ventriculography may also be carried out, if necessary. The great majority of subdural hematomas will be found through this approach. Some will be found only after passing a curved instrument into the subdural space, which may be done with ease in this region. When a semi-solid clot is present it can be evacuated after enlarging the subtemporal decompression. One of the obvious advantages of this approach is that if a large opening in the bone must be made to remove the clot the decompression is adequately protected by the temporal muscle and the fascia, which is not the case in the posterior parietal approach. Another advantage is the one already mentioned, that the solid portion of the clot most often lies in the temporal region and incidentally frequently covers the lateral portion of the temporal lobe to the base of the middle fossa. Clots in this latter region are not rare, as suggested in the literature. When exploration through the temporal approach fails to reveal a hematoma, it is obvious that additional burr holes may be indicated.

It was necessary to turn a bone flap in only 1 case in the series of 70, and this was to remove an organized liver-like clot, 31 days after injury.

SUMMARY

On clinical and pathological grounds, there is a group of patients who present the picture of an acute subdural hematoma and another group which may be classed as having a chronic subdural hematoma. In addition, there is a large number of individuals with subdural hematomas, who suffer from symptoms that are more correctly classed as subacute. Many of the patients in this latter group, although they remain drowsy or confused in some instances, have had time to recover, or largely recover, from their initial brain injury, but lapse into deepening drowsiness or show other signs of a severe progressive intracranial lesion in a week to several weeks after trauma. At operation, the hematomas in these individuals vary remarkably in structure, and pathologically, at times, more closely resemble the chronic than the acute type of lesion.

It is not with terminology that this paper is concerned, but rather with stressing the fact that many patients, suffering from large collections of blood in the subdural space, do not present an acute nor a chronic picture clinically or pathologically. Unless this is stressed the uninitiated may fail to be on the alert for the subdural hematoma that kills the patient at the end of the first week to several weeks after injury.

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THE USE OF DRY OXIDIZED CELLULOSE AS A PRIMARY HEMOSTATIC AGENT IN NEUROSURGERY

JOHN E. SCARFF, M.D., BYRON STOOKEY, M.D., AND FRANCISCO GARCIA, M.D.

Department of Neurosurgery, Columbia University, and Service of Neurological Surgery, Neurological Institute, Columbia-Presbyterian Medical Center, New York City

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OXIDIZED cellulose was first described by Yackel and Kenyon[11] of the Eastman Kodak Laboratories in 1941. This material consists of cellulose in the form of cotton, gauze, or paper, which has been subjected to oxidation by nitrous dioxide; as a result of this, carboxyl groups are formed, which render the material soluble in dilute alkali.

In 1943, Dr. Virginia Kneeland Frantz,[2] College of Physicians and Surgeons, Columbia University, published a description of experiments in which oxidized cellulose had been implanted in the abdominal muscles, brain, dura, joint cavities, and peritoneal cavity of dogs and cats. Dr. Frantz reported that in all but one of the experiments, the oxidized material had been completely absorbed within 4 weeks. Whether the disappearance was due to solution, or to digestion by phagocytes, she could not say. Tissue reaction at all stages was negligible. Proliferation of connective tissue and glia was minimal; no adhesions were found between dura and brain or within joint cavities.

Dr. Frantz and her associates[6] then reported further experiments in which they introduced packings of oxidized cellulose into surgical wounds in experimental animals. The second part of that paper included a report of 17 clinical cases contributed by various surgeons, in which control of bleeding had been achieved by the use of oxidized cotton.

In all of these experimental and clinical cases, the oxidized cellulose was used in the form of a surgical pack; its hemostatic effect was attributed entirely to the pressure exerted by the pack against the oozing surfaces.

A specific hemostatic property of oxidized cellulose was first cautiously suggested in a report by Frantz[3] in April of 1945, in which she stated, "in handling the soluble gauze it became apparent that it had *unexpected hemostatic action in its own right.*" This specific hemostatic property was repeatedly described in subsequent communications by these authors.[4,5,7] In all of these reports, they emphasized that this action was most effective when the material was used dry; when moistened, even with thrombin, its effectiveness was reduced.

It is important to distinguish between the use of oxidized cellulose as a primary hemostatic agent, and its use as an inert absorbable carrier for other recognized hemostatic agents such as thrombin. The use of oxidized cellulose as a mechanical carrier was first reported by Putnam[8] and subsequently by

Cronkite *et al.*[1] and by Uihlein *et al.*[9,10] None of these men was aware of the specific hemostatic properties of the oxidized cellulose.

Dry oxidized cellulose as a primary hemostatic agent in neurosurgery was first used at the Neurological Institute in April, 1944, by one of the authors (B.S.). Since that time, it has been used in approximately 2000 neurosurgical operations at this hospital. In none of these cases has the oxidized cellulose been used as accessory to another hemostatic agent.

Oxidized cellulose is available in two forms, cotton and gauze. The cotton is better adapted to neurosurgical technics. This material is packaged in such a way that secondary sterilization is unnecessary, rendering it immediately available to the surgeon at the operating table.

The greatest usefulness for this agent lies in the control of persistent oozing from broad surfaces. A thin layer of oxidized cellulose may be pressed against the under surface of bone in an osteoplastic flap to supplement waxing. Oozing from dural surfaces may be effectively controlled by placing small, thin pledgets of the oxidized cellulose directly over the bleeding point and holding it temporarily in place with a strip of dry cottonoid. Hemostatic action usually takes place within 30 seconds. Bleeding from Pacchionian granulations is effectively dealt with in this same manner. Small tears in the dural sinuses can likewise be controlled this way, although the texture of the material and its early absorbability render it unsatisfactory for the permanent repair of larger tears.

Small flat pledgets of oxidized cellulose may be applied directly to the brain itself to control bleeding from small vessels, either on the pial surface or from the raw surfaces remaining after cortical incisions. The method of application here is identical with that described for the dura.

In the sub-total removal of gliomata, intracapsular enucleation of tumors, and even in simple cavities left after the removal of encapsulated neoplasms, there often remain surfaces from which considerable oozing persists. Oxidized cellulose is particularly valuable in these situations. In many instances, the mere application of a thin sheet of the material against the oozing surface will suffice. In other cases where the bleeding is more brisk, the oxidized cellulose may be used as a surgical pack, in which the hemostatic effect of pressure is added to the specific hemostatic action of the cellulose. In this use, it has great advantage over other packs in that the bulk of the packing may be removed from the cavity after a few minutes, leaving a thin film affixed to the surface of the tissues to continue hemostasis. The removal of this type of pack does not cause fresh bleeding. The bleeding from hemangiomata affords a field of especial usefulness for this material.

In spinal surgery, oxidized cellulose is useful in controlling oozing from the lateral veins of the extradural plexus. Bleeding from small vessels on the surface of the spinal cord itself can be quickly controlled with small pledgets of the cellulose.

Bleeding from the stumps of peripheral nerves which have been resected and prepared for suture may be stopped by the application of small pledgets of the oxidized cellulose.

In general, oxidized cellulose will not stop bleeding from large or even medium-size arteries, nor is it an ideal material for repairing large tears in the dural sinuses, as mentioned above. In a few instances of emergency, however, it has been used with good effect for each of these purposes.

No undesirable side-effects have thus far been encountered in the use of oxidized cellulose. It produces no inflammatory reaction in the tissues, does not delay or impair wound healing, and when left in wounds does not produce foreign body reactions or discharging sinuses. Postoperative temperatures appear entirely unaffected by the presence of this material in wounds. No infections have been traced to it.

Oxidized cellulose develops no sensitivity by the tissues even after repeated use.

SUMMARY

1. The experimental and clinical development of dry oxidized cellulose as a primary hemostatic agent for neurosurgical procedures is briefly outlined.

2. Dry oxidized cellulose has been used as a specific primary hemostatic agent in approximately 2000 cases at the Neurological Institute of New York.

3. The specific hemostatic value of this material has been established.

4. Undesirable side-effects have not been observed.

5. Technics for use of dry oxidized cellulose in neurosurgery are described.

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CLASSIFICATION OF CRANIOSTENOSIS

DAVID FAIRMAN, M.D.,* AND GILBERT HORRAX, M.D.
Department of Neurosurgery, The Lahey Clinic, Boston, Massachusetts

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THE subject of this study is a discussion of the group of malformations in which there exists a premature synostosis of two or more membranous cranial bones. The history and pathologic anatomy of the condition will be considered and a new classification based on an anatomical, clinical and therapeutic standpoint will be suggested.

HISTORY

Homer described in the Iliad the ugliest man who came to Ilium saying that his head was peaked toward the top. Based on this Homeric reference, some French writers have used the phrase *Tête à la Thersite* for indicating oxycephaly.

Hippocrates[6] was cognizant of the cranial deformities and their relation to the cranial sutures. In the following excerpt he revealed to what extent these deformities were understood about 400 B.C.

> Men's heads are by no means all like to one another, nor are the sutures of the head of all men constructed in the same form. Thus, whoever has a prominence in the anterior part of the head, . . . in him the sutures of the head take the form of the Greek letter *tau*, T. . . . But whoever has the prominence in the back part of the head, in him the sutures are constructed in quite the opposite form to the former. . . . But whoever has a prominence of the head both before and behind, in him the sutures resemble the Greek letter *êta* H.

Although we were unable to find in the literature an interpretation of these statements formulated by Hippocrates, it is believed that when he spoke of the different forms of the cranial sutures he may have meant premature synostosis of the coronal or lambdoidal sutures in the first 2 cases, and closure of all the sutures including the sagittal suture in the 3rd case.

Oribasius, physician to the Emperor Julian, was aware of oxycephaly. Oribasius who, like Galen, was a native of Pergamos, wrote a digest of medicine and surgery in 70 books, of which 25 remain. He mentioned in some of these writings the cranial deformities associated with palatal defects and other abnormalities.

According to Greig[5] the term "oxycephalides" was used in 1830 by the great naturalist H. Milne Edwards, to designate a variety of crustaceans in which the head is more or less molded like a rostrum. Sommering in 1839, stated that an association appeared to exist between certain types of cranial deformity and the closure of some sutures.

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* Fellow in Neurosurgery, The Lahey Clinic, Boston, Massachusetts.

In 1851, Virchow presented a complete study of cranial deformities, giving for the first time a scientific explanation of these conditions. He established the relationship between the malformations of the skull and the premature synostosis of the cranial sutures, formulating the principle that, when synostosis of two bones occurs prematurely, normal growth is inhibited in a direction perpendicular to the obliterated suture line, and compensatory growth takes place in other directions.

Although Virchow failed to distinguish between chondrodystrophies and scaphocephaly or oxycephaly, his great merit consists in his observations that the premature synostosis of the sutures was the direct cause of the restriction of growth and, therefore, the primary cause of the cranial deformity, terming these conditions craniostenosis.

PATHOLOGIC ANATOMY

In order to understand the mechanism by which these conditions are produced, it is necessary to review the most important facts regarding the development of the skull in infancy.

From the embryologic standpoint, the cranium is primarily membranous. In the second month there appears a first and important differentiation. The inferior half is rapidly invaded by the chondrine and transformed into cartilage, while the superior part persists in the stage of simple membrane.

The first of these parts, the so-called chondrocranium, forms the base of the skull, that is, the ethmoid, sphenoid, and the inferior portions of the temporal and occipital bones. The second or membranous cranium constitutes the vault, that is, the frontal, parietal and occipital bones. The development of the base and the vault follows two different ways of ossification.

In the development of the base, there appear a number of points of ossification in the cartilaginous tissue. The ossification is not complete until the age of 6 or 7 years. Development of the vault is more complicated. According to Ford[3] the membranous bones of the vault are still slightly separated at birth although their margins are in apposition, but no bony union exists, as may easily be determined by palpation. The anterior and posterior fontanelles are still evident. Ford stated that between the 6th month and the end of the 1st year the sutures of the vault become closed. This closure of sutures is apparent because the serrated margins begin to interlock, although x-ray examination reveals that the bones are not actually fused. The posterior fontanelle closes, under normal conditions, by the 2nd month and the anterior fontanelle closes between the 14th and the 22nd month.

During childhood the margins of the cranial bones are not actually fused; therefore, it is still possible for the skull to grow. The sutures are interlocked so firmly, however, at the age of 10 years, that increased intracranial pressure causes little or no enlargement of the head.

The sutures play a very important role in the development of the skull. When the ossification invades the suture and the different bones of the

vault become actually fused, then the synostosis is physiologic, when it begins during middle age. Testut[8] said that the physiologic synostosis begins at 40 or 45 years of age and is not complete until the age of 75 or 80. In the various conditions under consideration, the cranial bones are united prematurely, becoming fused before birth. The line of union disappears completely or is marked by a bony ridge. This process constitutes the pathologic synostosis which is of practical significance only when it takes place during intrauterine life or in the 1st year of postfetal life. During this period, growth of the skull is possible on account of the suture spaces. Later the skull develops only by periosteal apposition and resorption, just as in the basal synchondrosis.

The earlier the pathologic synostosis takes place, the more marked is the degree of skull contraction or craniostenosis. On the other hand, closure of a single suture may cause only some contraction since those sutures that remain open take on a compensatory function. It is possible to see externally this compensation, revealed in the irregular development of the skull in various directions.

PATHOGENESIS OF CRANIOSTENOSIS

In 1856 Minchin, from Dublin, in a paper based on the fact that there is often a thickening in the region of the obliterated suture, conceived the idea of a single parietal bone extending bilaterally from the medially sutured center. He thought that the thickening above mentioned constituted a single center of ossification for two bones instead of the usual two different centers developing two separate bones. It is impossible to maintain this theory due to the fact that the thickening always shows, from the histologic standpoint, remnants of the early suture.

Fournier[4] stated that syphilis was the etiologic factor in the cranial malformations under discussion, but in all cases in which Wassermann tests were done they were negative and there were no pathologic conditions indicating congenital syphilis.

Virchow, in his fundamental work already mentioned, described an adult oxycephalic skull showing an inflammatory process, involving one of the cranial bones partially. Based on this evidence only, he conceived the idea that an inflammatory process developed in the meninges, and later involved the cranial bones. Therefore, their capacity for further marginal growth was inhibited, producing the pathologic suture.

The main objection to Virchow's thesis consists in the fact that no one so far has been able to demonstrate macroscopically or microscopically an inflammatory process in the lines of the sutures in postfetal life.

Thoma,[9] in a series of articles published in *Virchow's Archives* between 1907 and 1918, formulated the hypothesis that external pressure produced all pathologic closure of sutures during fetal life. He meant by external pressure the compressing mechanism of the uterine musculature.

Since the pressure exerted by the uterus under normal conditions is a fluid pressure, we must assume that Thoma believed that oligohydramnios

was always present. However, Park and Powers,[7] in their classical work published in 1920, demonstrated that in their series of 25 cases a history of oligohydramnios was not present in any instance. On the contrary, they reported in 3 cases a history of polyhydramnios. Therefore, Thoma's theory is unacceptable.

Rieping, in 1919, expressed the theory that a defect in the blastemal stage of cranial development was responsible for the displacement of the primary ossification points of the cranial membranous bones toward the suture lines. He felt that when the primary ossification centers of the parietal bones were dislocated toward the midline, the result would be a premature synostosis of the sagittal suture. The same principle applied to the frontal and occipital bones would explain the other varieties of craniostenosis. It is interesting that the same hypothesis was formulated as early as 1875 by Morselli.

Park and Powers insisted on the fact that the margins of the cranial bones were kept apart by interstitial normal growth of mesenchymal tissue. Due to a defect in the germ plasm the capacity of normal growth of the mesenchyme is diminished or lost, without developing the exudate enabling it to resist ossification, thus explaining the premature synostosis.

Undoubtedly, the theory of Rieping, and of Morselli, complemented by that of Park and Powers, resists any fundamental criticism, being so far the most logical hypothesis acceptable.

CLASSIFICATION AND CLINICAL SYMPTOMS

The only formal classification of craniostenosis has been made by Greig,[5] who considered oxycephaly as a whole and divided it into three types:

I. *True oxycephaly* which presents a general craniofacial stenosis. This is congenital and often associated with syndactylism or other deformities of the extremities.

II. *Delayed oxycephaly* which may appear at any time during childhood and never presents other deformities.

III. *False oxycephaly* which is localized synostosis, not congenital, and is often of definite origin.

Analyzing the original papers published by Greig in 1926, we find the following facts: *first* that his conception of true oxycephaly was based on 3 patients, aged 58, 58 and 45 years respectively. The 3 patients presented deformity of the head and symptoms of mental deficiency, with 2 of them showing deformities of the extremities. In spite of the fact that these 3 cases did not present any x-ray evidence, we know that a synostosis discovered after 45 years of age cannot be considered unusual and it is impossible to make a retrospective diagnosis to establish at what age closure of the suture occurred. Furthermore, we believe that it is impossible to make a diagnosis of oxycephaly based on the shape of the deformity of the head and the neurologic symptoms only. It is indispensable to have x-ray evidence of the premature closure of the sutures.

As far as the deformities of the extremities are concerned, we believe it

is unjustifiable to regard the combination of malformations of head and extremities as a clinical entity. The significance of their occurrence together appears to be merely an inconstant association, since there is a special tendency when malformations appear in the head to appear also in the extremities.

The *second* point is that Greig's conception of delayed oxycephaly is based on 2 patients aged 8 and 9 years respectively. He thought that in these cases the condition was not congenital. The deformity was not exaggerated and did not present any related concomitant somatic defect. No roentgenograms of the skull were taken of one patient. In the other patient, although there was an x-ray report of digital impressions over the cranial vault, no mention of synostosis was made. For these reasons it is doubtful whether a diagnosis of oxycephaly can be accepted.

Greig's *third* point is a classification of his conception of false oxycephaly based on the examination of skulls in the museum of Edinburgh (all of them middle age or older). He stated that in false oxycephaly the synostosis was limited to a few or even to one suture, that facial sutures never participated and that no association of deformities of the extremities coexisted. He stated that the base of the skull was not affected and that the condition was not congenital. Furthermore, he stated that the deformity of the head in false oxycephaly was a misleading feature and he did not recognize this group as a pathologic entity.

We believe that Greig's description of false oxycephaly fits perfectly the present classification of brachycephaly and scaphocephaly in which, as we know, partial synostosis is responsible for the clinical symptoms.

Since Greig published his article in 1926, his classification has been unanimously accepted and followed, up to the present time. In reviewing the literature no criticism of Greig's classification has been disclosed. On the contrary, his classification is followed carefully, which in our opinion is misleading and adds to confusion for the reasons previously cited.

Based on anatomical and clinical facts, the following classification is suggested.

- Craniostenosis
  - Incomplete
    - Scaphocephaly
      - Compensated
      - Decompensated
    - Brachycephaly
      - Compensated
      - Decompensated
  - Complete
    - Oxycephaly
      - Compensated
      - Decompensated

As may be seen in this classification, we consider the anatomical deformity of the head and the neurologic symptoms. Deformity of the skull is a result of Virchow's law: limitation of expansion in directions perpendicular

to the suture-line closure causes overexpansion to take place in the regions where the sutures remain open.

The *first* is scaphocephaly involving closure of the sagittal suture with compensatory lengthening of the skull in its anteroposterior dimension and often widening and elevation of the frontal region.

The *second* is brachycephaly involving synostosis of the coronal sutures, presenting normal growth in the anteroposterior direction. The skull becomes broad and short; the root of the nose is also widened so that the eyes are placed very far apart. This condition also has been termed acrobrachycephaly, but the prefix "acro" has been applied to oxycephaly as well, creating a misnomer.

The *third*, oxycephaly, involves the coronal and the sagittal sutures and prevents normal expansion both in the transverse and anteroposterior directions. Other sutures may be involved as well; the skull can expand only in the vertical direction. This condition has been termed turricephaly, and acrocephaly also.

In our opinion, therefore, the above terms, namely, oxycephaly, brachycephaly and scaphocephaly, represent the only logical nomenclature for the various types of craniostenosis.

The neurologic symptoms are dependent upon the degree of contraction of the skull. The craniostenosis is decompensated when the patient develops chronic increased intracranial pressure as evidenced by headache, vomiting, papilledema, failing vision and convulsive seizures. Optic atrophy with blindness and psychic disturbances may also occur.

An extremely striking feature of craniostenosis is bilateral exophthalmos, especially in oxycephaly and scaphocephaly. In well developed cases the protrusion of the eyeball may be so severe that it is impossible for the patient to close the lids and is frequently associated with strabismus. The exophthalmos may be due to the fact that the orbits are very shallow and oblique with the anterior and middle fossae deeper than usual and shortened in the anteroposterior diameter.

It is impossible from the neurologic symptoms to make a differential diagnosis between the varieties of craniostenosis, the difference being based only on the degree of deformity of the head correlated with the roentgenographic findings.

Craniostenosis is compensated when a compensatory growth of the skull occurs at the open sutures so that symptoms of increased intracranial pressure do not develop.

The x-ray examination enables us to determine with accuracy the variety of craniostenosis and the degree of contraction of the skull. In oxycephaly the coronal and sagittal sutures are seen to be synostosed. It is essential in this condition that the premature synostosis of the sutures above mentioned be present, although the lambdoidal suture may be involved also.

In brachycephaly the coronal suture is synostosed more or less completely but the sagittal and lambdoidal sutures remain open in many cases long after they should be closed.

Finally in scaphocephaly the characteristic feature from the x-ray standpoint is the premature closure of the sagittal suture while the coronal suture may be widened.

DIFFERENTIAL DIAGNOSIS

We have excluded from this study the craniofacial dysostosis of Crouzon, included by some authors, because it is very rare. In addition, the description of this condition given by Crouzon[1] in 1929 is not clear enough to consider it as a form of craniostenosis.

As far as chondrodystrophy is concerned, included by Virchow in the group of craniostenosis, it is now definitely identified as a different pathologic entity. This condition is due to a premature union of the bones which form the base of the skull. The defective development involves not only the bones of the base of the skull, which are, as we know, of cartilaginous origin but also affects practically all the other cartilaginous bones, producing among other symptoms shortening of the arms and legs. The enlarged head seen in chondrodystrophy is produced, in some cases at least, by hydrocephalus. Dandy,[2] who demonstrated by air injection in 1921 the existence of hydrocephalus in this condition, suggested that it might be due to the shortened base of the skull which causes bending of the aqueduct or obstruction of the cisterna magna, at least during early life and later corrected.

SUMMARY

Deformities of the head which are now designated as craniostenosis were known to the ancient Greeks and were described by Hippocrates in 400 B.C. The various explanations of the pathogenesis of this condition since Virchow's original study have been discussed and analyzed. Based on anatomical and clinical grounds a new classification of the forms of craniostenosis is offered.

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ARNOLD-CHIARI DEFORMITY WITHOUT BONY ANOMALIES

HOMER S. SWANSON, M.D., AND EDGAR F. FINCHER, M.D.
Department of Surgery (Neurological), Emory University, Atlanta, Georgia

(Received for publication November 13, 1948)

SINCE the early and original contributions of Arnold[3] and Chiari[5] on the malformation that now bears their names, numerous articles have appeared pertaining to the mechanism, diagnosis and treatment of this anomaly. The occurrence of this deformity has usually been found in infants to be associated with extreme degrees of spina bifida and cranium bifidum. When encountered in adults this pathologic entity has generally been seen in conjunction with some type of bony anomaly of the cranio-vertebral junction, such as basilar impression (platybasia) or the Klippel-Feil deformity.

A survey of the literature, however, reveals several cases in which this malformation has been proven either at operation or autopsy, to exist without spina bifida or coexisting bony anomalies of the spine. The case reported here illustrates the fact that this anomaly may exist in the absence of either bony deformities or spina bifida, and furthermore that it may occur in the very young, our patient being the youngest of those recorded. It is because of this fact and in view of the unusual progress and diagnostic difficulties presented by this case that we set forth our experiences in dealing with this problem of the Arnold-Chiari malformation without demonstrable bony anomaly.

CASE REPORT

History. A 5-year-old white female was seen in consultation on April 28, 1947. She was the first born of the family. Her birth and developmental history were entirely normal, and she had had none of the usual childhood diseases. In November 1946, the mother first became aware of the fact that the child was less active than usual, that she appeared listless and preferred not to run and play as she had formerly. She offered no specific complaints other than that her head ached and that exercise caused pain in her neck. By December 1946 such general lassitude and weakness of the lower extremities had developed that her schooling was discontinued. It was then that the mother noted a change in the child's gait, described as a tendency to drag her feet when walking. Following tonsillectomy in February 1947 there was noted for the first time a weakness in the upper extremities similar to that seen in the lower. In the latter part of March, 1947 the child had to be confined to bed because of profound weakness of all extremities. She was unable to elevate her arms above the shoulder level. She could not hold her head erect nor was she able to hold her body erect upon attempting to stand upright. Two weeks before the initial examination the child had, on two occasions, regurgitated liquids through her nose. At about this same time there developed huskiness of her voice. This illness had not been attended by fever; she had not complained of any specific pain and had not experienced any cranial nerve disturbances other than the dysphagia.

Neurological Examination. The appearance of this child was similar to that of an adult in an advanced stage of myasthenia gravis. She was carried into the examining room by her father, and when placed in the erect position she immediately sagged against the support of his arms, the knees flexed and were adducted, the arms hung limply by her side, and the chin rested against the chest. When she attempted to look at the examiner she did so by elevating her eyes and lids but not her head. She breathed through her mouth with her tongue partially protruded, and there was a salivary drooling. Her breathing was rapid and labored and was performed entirely by the intercostal muscles. When held in an upright position she was able to offer some support of herself on her toes and heels. The handgrips and finger movements were likewise weak. Individual muscle tests revealed that the most marked handicaps were apparently confined to the group supplied by the upper and middle cervical cord. There were no fibrillary twitchings noted and the only demonstrable muscle atrophy was confined to the intrinsic muscles of the left hand. This produced a modified ulnar type of deformity. There was no involuntary nuchal rigidity, but she complained of pain when her head was moved.

Examination of the vertebral column revealed no palpable deformities of the cervical spine, and there were no findings to suggest a spina bifida at a lower level. The cranial nerve study was not noteworthy aside from obvious palate and tongue weakness which was not attended by atrophy or fibrillary twitching of the tongue. The sensory examination was entirely normal for all modalities. There were no abnormal cerebellar objective evidences. Reflex studies elicited brisk triceps responses bilaterally, but the biceps reflexes and the abdominal reflexes were absent. There was a marked exaggeration of the knee and ankle reflexes. Transient ankle clonus was present and the Babinski reflex was bilaterally positive.

Course in Hospital. The child was immediately hospitalized with a presumptive diagnosis of an infiltrating tumor of the upper cervical spinal cord and medulla oblongata. Roentgenograms of the spine revealed no evidence of a congenital anomaly. There were no suggestions of a widening of the interpedicular measurements, laminal erosions or traumatic damages; nor did X-rays of the skull depict abnormalities. A lumbar puncture was attempted and although the operator was certain the needle was within the canal, clear fluid was obtained only by resorting to abdominal compression, and the CSF pressure was recorded at 30 mm. of water. Fluid sufficient for a protein determination was finally removed and 1 cc. of lipiodol was then introduced. The protein was reported as 63 mg. per cent, the cell count 0. An attempt was made to fluoroscope the movement of the lipiodol but it was found to be immobile, apparently having been introduced into the subdural space. A 2nd attempt was made to determine the CSF dynamics, with similar results except that on the 2nd occasion jugular compression did increase the flow of the spinal fluid measurably.

At this point a neurological consultant confirmed the previous findings and corroborated the probable diagnosis of glioma of the brain stem and cervical cord. A trial of roentgen therapy was agreed upon, inasmuch as there had been a distinct improvement clinically. Even before changes secondary to the X-ray therapy could be expected, the child became more active, was able to engage in minor activities on the ward and no longer exhibited the positive Babinski reflexes. At the completion of the course of roentgen ray therapy she was ambulant with increasing power, no longer exhibited difficulty in swallowing or breathing, and could elevate her arms above her head. She was discharged from the hospital to be observed at frequent intervals.

Subsequent Course. The patient was seen at frequent intervals during the next 3 months, and at each visit the picture presented was dissimilar. On one occasion she had reverted to the condition seen on the first examination, with the added finding of nystagmus on lateral gaze. On another occasion she appeared bright, sufficiently strong to walk unaided, and presented no very positive neurologic findings. During one of her more marked periods of regression, she was re-admitted for further CSF study. The manometric studies were entirely normal, and the protein was reported as 54 mg. per cent. A trial of prostigmine was given without beneficial effect and she was discharged definitely improved, this improvement lasting 1 month.

For a short period following her return home she improved markedly, to the extent that her parents considered sending her to school. Within 1 month, however, her condition became progressively worse. In addition to the original symptoms and the nystagmus on lateral gaze, there was paresis of the tongue and palate movements with complete areflexia in the upper extremities. There developed disturbing paroxysms of coughing due to swallowing difficulties. She was readmitted to the hospital for the 3rd time Nov. 15, 1947 for encephalography. This procedure was elected because of the technical difficulties encountered previously with the lipiodol studies and in the hope of visualizing the upper cervical and lower cranial spinal fluid pathways.

Encephalogram Report. With the patient under pentothal anesthesia and in an upright position, a lumbar puncture was performed. Fractionally, after considerable time had elapsed, a total of 20 cc. of clear fluid, containing 30 mg. per cent protein, was obtained, and 25 cc. of oxygen introduced. Routine skull roentgenograms as well as stereoscopic views of the cervical spine were obtained with the patient semi-erect on the table. There were only a few small gas shadows over the convexity of the brain, but none visualized in any of the ventricular cavities. On the cervical films the air column was seen to extend as high as the 2nd cervical level, at which point it appeared to be cut off. For a distance of 2 spinal segments below this point the column was distinctly reduced in width. The findings suggested an Arnold-Chiari deformity, and an exploratory operation was advised.

Operation. On Nov. 18, 1947, under intratracheal ether anesthetic, a combined suboccipital craniotomy and upper cervical laminectomy was carried out through a midline incision extending from the greater occipital protruberance to the level of the 4th spinous process. As the cervical arches were exposed, an anomalous, tremendously dilated and tortuous vein, apparently draining into the posterior internal vertebral venous plexus, was encountered. This vein lay over the 1st laminal arch, on the left, and after hemostasis of this large vessel an almost complete erosion of this arch was noted. This was in sharp contrast to the normal-appearing 1st arch on the right.

Following the removal of the 1st and 2nd cervical spinous processes and arches, a wide, firm fibrous band, which appeared to be a reduplication of the posterior occipito-atlantal ligament, was noted. This extended from the upper level of the 1st arch to the midportion of the 2nd, and measured 3 mm. in width. After resecting this tough ligamentous band, a deep transverse notch was noted in the dura overlying the greater cistern. Freeing this constricting band did not result in reexpansion of the dura. Accordingly, the right posterior horn of the lateral ventricle was tapped and no increased ventricular pressure was noted nor did evacuating the fluid from this cerebral cavity improve the dural deformity.

The bone over the suboccipital region having already been removed, the dura was opened over both cerebellar hemispheres and as far inferiorly as the level of the

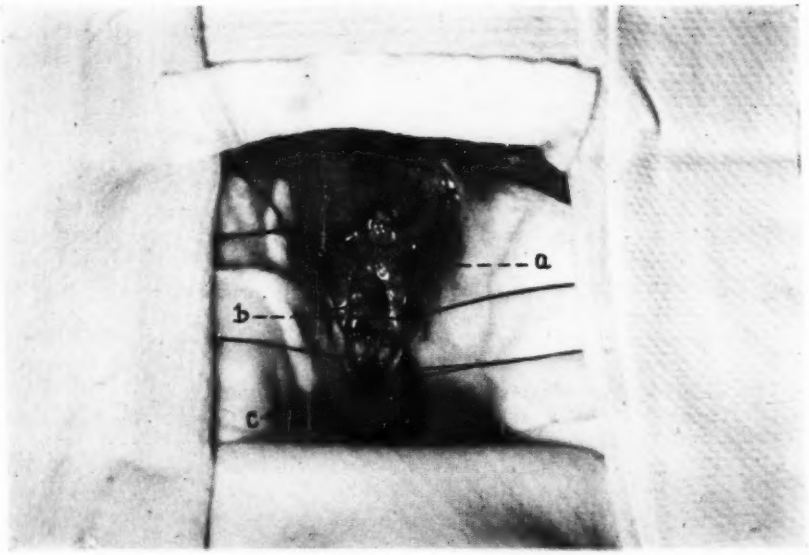

FIG. 1. Photograph of operative findings. (a) Occipital-cervical junction. (b) Caudally displaced and distorted cerebellar tonsils. (c) Third cervical level of spinal cord.

3rd cervical arch. This exposure revealed both cerebellar tonsils displaced caudally into the cervical canal to the level of the midportion of the 3rd cervical arch (Fig. 1). Both tonsils were literally glued together by thin arachnoidal adhesions which were easily separated. Both inferior cerebellar tips were yellow and atrophic in appearance. These structures were flattened to no more than 1 mm. thickness at their caudal ending. With the freeing of the arachnoidal adhesions and separation of the tonsils, there then occurred a brisk escape of clear fluid from the 4th ventricle. By retracting and separating the cerebellar tonsils, an adequate visualization of the 4th ventricle was afforded and it was noted that the lower portion of the 4th ventricle was elongated and displaced downward into the vertebral canal (Type II, Chiari).

Inspection of the site of junction of the medulla oblongata and the cervical cord disclosed a small area of cystic degeneration in the midline and a distinct cleft in the cervical cord corresponding to the level of the similar indenture noted in the dura. This cystic area corresponded to the lowermost portion of the 4th ventricle. Both the 1st and 2nd cervical nerves were observed to ascend obliquely to their exit from the spinal canal rather than to run in their usual transverse direction.

Inspection and palpation of the cerebellar hemispheres disclosed no unusual findings, and exploration of the cerebellopontine angles revealed no tumor. At this point, however, the explanation for the peculiar appearance of the cerebellar hemispheres became apparent: there was no structure present corresponding to the vermis. With a continuous escape of fluid from the 4th ventricle, it appeared obvious that no obstruction of the aqueduct existed.

Freeing the obstruction at the level of the greater cistern required removal of the 3rd cervical arch and amputation of both cerebellar tonsils. This procedure effected an adequate decompression, and the operation was terminated by a partial approximation of the dura over the cervical cord and cerebellar hemispheres, leaving that cistern open. The wound was meticulously closed in layers to prevent a cerebrospinal fistula, and the patient was returned to her room in excellent condition.

Postoperative Course. The course following surgery was uneventful. On the 1st postoperative day a spinal puncture was done to ascertain if a free communication of cerebrospinal fluid had been accomplished. It apparently had, as 20 cc. of slightly bloody fluid was removed. By the 3rd postoperative day the child was able to swallow liquids without difficulty, and our earlier fears of a respiratory catastrophe were relieved. The nystagmus and pathologic reflexes were not noted after the 4th day. When allowed up on the 6th day, the child was able to hold her head erect, to elevate her arms above her head, and to walk unaided, though she did so with a wide base. Upon discharge from the hospital on the 10th postoperative day she was able to dress herself, to walk unassisted, and was eating solid foods without regurgitation.

When last seen 1 year after operation, the child had improved markedly. She had regained full motor power and coordination, no longer complained of any discomforts and had been able to engage in the physical activities of her schoolmates without limitation. The only positive neurologic findings consisted of persisting areflexia in the upper extremities and hyper-reflexia in the lower extremities without pathologic toe signs. There was still persisting evidence of minor atrophies of the left hand, but the handgrip and finger movements were strong.

DISCUSSION

Many authors in discussing the Arnold-Chiari malformation have stressed the combination of the Chiari deformity and the usually described

bony lesions, pointing out the necessary accompaniment of the latter in order to conform to the Arnold-Chiari syndrome. In 1938, McConnell and Parker[7] reported 5 cases of the Arnold-Chiari malformation. The youngest patient was 10 years of age, the oldest 32. In none of these patients was there demonstrable evidence of a spina bifida deformity, but all had an associated internal hydrocephalus. No mention was made by these authors of cervical spine pathology. In the same year, Aring[2] reported a single case in an adult without vertebral changes, and suggested that his case might represent a variant of a congenital spinocerebellar ataxia. Adams, Schatzki and Scoville[1] reported 2 instances of this malformation, 1 in a 17-year-old boy without demonstrable evidences of a spina bifida, and the 2nd in a 13-year-old boy who had had a myelomeningocele repaired at birth. Similarly Penfield and Coburn[9] have recorded a case of a 29-year-old woman in whom symptoms due to this anomaly did not appear until 26 years after the repair of a dorsal myelomeningocele. These 2 cases illustrate the latent period of development of symptoms which may follow repair of a spina bifida and support the theory of Russell[10] and others that the Arnold-Chiari malformation may develop in association with a spina bifida. This deformity may occur late in life without demonstrable cause, as evidenced by the case reported by Bucy and Lichtenstein.[4] In this instance, a 40-year-old negress without demonstrable bone pathology or cause for the lesion was found to have the classical Arnold-Chiari deformity. Ogryzlo[8] reported 3 adults, operated upon by Dr. McKenzie, who had this anomaly yet did not present vertebral changes. In 2 instances a preoperative diagnosis of congenital herniation of the cerebellum had been made.

Most authors have stressed the fact that an internal hydrocephalus accompanies this malformation, and some have expressed the belief that the hydrocephalus may be responsible for the deformity. In the case reported here, even though clinical evidence was present intermittently, and even though at operation there appeared a complete block of the cerebrospinal fluid at the cisterna magna, a hydrocephalus from ventricular estimation was not present. Although increased intracranial pressure may produce the so-called cerebellar pressure cone, the extreme prolapse to the degree frequently encountered in this malformation is rarely seen in the usual hydrocephalus.

The Arnold-Chiari malformation is apparently a congenital deformity and may be associated with a wide variety of other anomalies in addition to the bony deformity. The incomplete development of the cerebellar vermis has repeatedly been encountered, as in the case reported here. In Case V of McConnell and Parker, there was a congenital absence of the foramina of Magendie and Luschka. Lichtenstein[6] reported an associated aqueductal stenosis.

The etiology of the malformation in the presence of spina bifida has apparently been well established as due to the traction force exerted against the medulla and cerebellar tonsils with fixation of the spinal cord at the site

of the spina bifida. With continued elongation of the vertebral column, these structures are pulled downward through the foramen magnum. As Bucy and Lichtenstein have suggested, this same traction force may produce the malformation in the absence of firm fixation of the brain in the cranial cavity. This latter supposition has not been confirmed by anatomic studies. The Arnold-Chiari malformation therefore may occur in association with a number of other congenital anomalies and need not in each instance be combined with either a spina bifida or other cranio-vertebral deformities.

SUMMARY

1. A case of Arnold-Chiari malformation without demonstrable spina bifida, hydrocephalus or other cranio-vertebral anomalies is reported.

2. Attention is called to the fact that this malformation may occur in association with a number of congenital anomalies and need not in each instance be combined with spina bifida, hydrocephalus or cranio-vertebral deformities.

3. This malformation should be considered in evaluating acquired pyramidal tract disturbances.

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A NOTE ON IDENTIFICATION OF THE MOTOR SUPPLY TO THE DETRUSOR

DURING ANTERIOR DORSOLUMBAR RHIZOTOMY

ROBERT A. KUHN, M.D.*

Department of Physiology, Johns Hopkins University School of Medicine, Baltimore, Maryland

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DURING recent years surgical treatment of muscle spasm below the region of severe spinal cord injury has received considerable impetus. In 1933 Munro[8] attempted bilateral intradural division of the lower thoracic, lumbar, and 1st sacral anterior nerve roots in markedly spastic patients with anatomic transection of the spinal cord, with the intention of converting their paralysis into a flaccid one, and enabling otherwise impossible rehabilitation to be carried out. In 1945 the same author described a revised surgical technique for anterior dorsolumbar rhizotomy, and reported a total of 10 civilian patients treated in this manner.

Among the numerous casualties with spinal cord injuries in World War II were many men in whom there developed extreme involuntary activity in the skeletal musculature below the level of their cord lesion.[4,6] Medical therapy of this reflex activity proved discouraging.[5] Botterell *et al.*[1] reported a series of 5 anterior rhizotomies for "mass reflex" in 1946, and shortly thereafter Freeman and Heimburger[2] described operative technique and results of rhizotomy in 15 paraplegic men, with observations on an additional 13 subjects operated upon by other surgeons. Invariably, reports on this operative procedure stressed the difficulties inherent, during surgery, in identifying the exact segmental level of a given anterior root. In his first cases Munro counted upward from the 5th sacral anterior root, but soon discarded this method as unsatisfactory, and utilized instead the last firm dentate ligament as an indication of the level of the 1st lumbar root. Macdonald *et al.*[7] pointed out, however, that the last dentate ligament is relatively variable in level, and does not have a constant relation to the 1st lumbar root. Other investigators[2] stated that, though the last firm dentate ligament is a fairly constant finding at the level of the 12th dorsal vertebra, anterior roots below this level may appear to be fused, and that resultant miscount during rhizotomy would result in section of motor roots to the bladder. The same authors suggested that an anterior root too large to be enclosed in a single silver clip should be designated as two roots.

It appears evident that, even with extreme care in the visualization and identification of anterior roots by all available methods, anterior rhizotomy performed in this manner is attended by considerable hazard to the motor

* Formerly, Chief of Surgery, Paraplegia Service, Cushing V. A. Hospital, Framingham, Massachusetts.

supply of the detrusor. Accordingly, a method of more positive identification of the sacral roots constituting the efferent innervation of the bladder was utilized.

Anterior dorsolumbar rhizotomies were performed by the author during the years 1946–1947 on 22 patients exhibiting severe mass extension or mass flexion reflex activity below the level of cord injury. Indications for operation were similar to those described by other authors.[2,8] Exploratory laminectomy to verify the completeness of cord interruption was performed in the majority of the subjects. Intravenous sodium pentothal was utilized in approximately $\frac{1}{2}$ of the cases as the anesthetic of choice. Other men were operated upon with no anesthetic, or with local infiltration of 1 per cent procaine hydrochloride containing epinephrine. The difficulties in identification of sacral motor roots innervating the bladder were largely obviated by reliance on the effects produced on the musculature of the bladder through electrical stimulation of isolated ventral roots.

METHOD

The day prior to surgery, a Foley urethral catheter was inserted in the bladder of the patient, and a preoperative cystometrogram performed. The rate of introduction of fluid into the bladder was not allowed to exceed 90 drops per minute, and a curve of all (except respiratory) fluctuations in bladder pressure was plotted. The volume at which the first maximal detrusor contractions occurred was thus determined. Under standard conditions in a given subject, this critical volume did not vary more than 1–2 oz., hence one preoperative cystometric determination usually sufficed.

In the operating room, the patient was placed prone on the table with the latter broken at the hips to flatten the normal lumbar lordosis. The thighs were thus placed in a position of partial flexion, effectively minimizing flexor movements of the lower limbs even in those patients showing intense reflex activity below the level of cord transection. It was not necessary to adopt the lumbar puncture position, despite moderate contractures at the hips in a few men. Heavy towel rolls were now placed beneath each inguinal crease to elevate the hips of the patient and, in addition, to insure that no portion of the ventral abdominal wall was in contact with the table. The urethral catheter was led down between the thighs and attached to a modified Munro tidal-drainage unit,[9] allowing an ample length of rubber tubing at the perineum. The patient was then draped in the usual manner, and the entire apparatus shielded from the operative field.

Standard midline incision for exposure of the lower end of the cord was carried out, and the medial ends of the last dentate ligaments used for retraction of the conus and cauda equina. Immediately prior to root stimulations, an assistant drained the contents of the bladder, and introduced slowly a volume of saline slightly less than that previously estimated to be necessary for reflex detrusor contraction.

Root stimulation was now carried out, beginning with the root exiting at the D11–D12 intravertebral foramen and proceeding caudally, each root being lifted with a nerve hook, blocked from the field, and stimulated at 1–2 volts for 5 seconds. A Bovie stimulator was used in a few instances, but a later model Garceau peripheral nerve stimulator was employed in the majority of operations. All anterior roots

which, when stimulated, caused skeletal muscle contraction and failed to result in a specific type of detrusor activity were severed. The abrupt contraction of large skeletal muscle groups resulting from electrical stimulation was accompanied by a short, sharp rise and fall of the fluid column, resulting probably from mechanical alteration of intra-abdominal pressure. Pressure changes that apparently represented contraction of the detrusor were distinguished easily. The latter typically showed an initially slow but steadily accelerating rise in bladder pressure to heights determined by the particular nerve stimulated, but invariably to a level far in excess of the alteration evoked by mechanical disturbance. The decline in pressure at cessation of stimulation usually occurred in a series of steps, the fluid column stabilizing momentarily at each new level until the normal baseline was reached.

Stimulation of the lower thoracic, all of the lumbar, and the 1st sacral anterior roots never resulted in typical detrusor activity, and stimulation of large sacral roots (i.e., S1 and S2) rarely elicited response. By count from the last firm dentate ligament, the 3rd and 4th sacral anterior roots yielded maximal detrusor contractions in the majority of the men. It appeared that the size of the sacral root was more indicative of its relation to the bladder than was the apparent segmental level as determined by counting from above. If the 2nd sacral anterior root elicited detrusor activity, that root was invariably much smaller than the anterior root immediately above. Stimulation of an occasional barely visible anterior rootlet arising near the filum terminale resulted in maximal contractions to 80–100 cm. of water.

RESULTS

In 22 patients, preoperative cystometrograms showed 13 with hypertonic type reflex bladders. Of these 13 men, 11 displayed, after anterior rhizotomy, amelioration of excessive detrusor activity sufficient to reclassify the bladder to that of reflex type. The bladder status remained unchanged in 2 subjects of this group. Four of the 9 other patients exhibited autonomous bladder activity prior to rhizotomy; in 3 men activity was unchanged, and 1 developed an excellent reflex bladder after rhizotomy. Four subjects possessed reflex bladders before operation, 2 of them promptly regaining original reflex bladder activity after surgery, and 2 manifesting autonomous reflex activity 2 months after rhizotomy had been performed.

COMMENT

Available studies indicate that the motor fibers to the detrusor and those to the external urinary sphincter travel through the same sacral roots. It is therefore improbable that a sacral root which, when stimulated, fails to cause detrusor contraction, will carry fibers essential to reflex control of the external urinary sphincter. Heimburger and his co-workers,[3] using peripheral procaine infiltration, found that the 3rd sacral root was the predominant one supplying motor innervation to the detrusor, while Shelden and Bors[10] referring to unpublished data, state that the 3rd sacral root is chiefly responsible for contraction of the urinary sphincter.

SUMMARY AND CONCLUSIONS

1. A method for more accurate identification of the motor supply to the detrusor during anterior dorsolumbar rhizotomy is described.

2. Electrical stimulation of the 3rd and 4th sacral roots, during 22 such operations, caused maximal detrusor activity in the majority of the patients.

3. In view of the safety and relative ease of this method of sacral root identification, it is suggested that the utilization of subarachnoid alcohol block for abolition of extreme reflex muscular activity below a cord lesion be restricted to only those patients displaying total lack of detrusor innervation as determined by cystometric studies.

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DUPLICATION OF THE SPINAL CORD (DIPLOMYELIA)

AN ACCOUNT OF A CLINICAL EXAMPLE WITH A CONSIDERATION OF OTHER REPORTS*

WILFRED PICKLES, M.D.

Department of Neurosurgery, Rhode Island Hospital, Providence, Rhode Island

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On Dec. 3, 1946, a 14-year-old girl (B. P., R. I. H. 414450) was admitted to the Neurosurgical Department of the Rhode Island Hospital, complaining of pain and numbness in the right leg and foot, with disability in walking. Her family history was not remarkable. She had had an enlarged thymus, scurvy, and cyclic vomiting as an infant. At 10 years of age she began to limp and walked with the right foot turned in.

FIG. 1. Pantopaque myelogram of the author's patient.

Examination showed a small, short right foot with high arch and persistent inversion, with atrophy and general weakness of the muscles of the entire right leg; there was anesthesia to light touch on all the toes on the right, with hypesthesia to touch and pinprick up to the knee, and normal temperature sense. Reflex findings were normal except for an absent knee jerk and a sluggish ankle jerk on the right. Roentgenograms of the skull and right foot were normal; the spine showed flattening and deformity of the 10th, 11th and 12th dorsal bodies, with narrowing of the interspaces, partial absence of the 11th and 12th dorsal neural arches, and complete absence of the 1st lumbar arch. Lumbar puncture findings were normal except for a protein content of 62 mg. per cent.

Pantopaque myelography (Fig. 1) was done by Dr. Thomas McOsker; and Dr. Lawrence Martineau, the roentgenologist, reported a split in the oil column at the level of the congenital defect, with a large irregular defect at the 10th dorsal body without complete obstruction; he felt that this appearance was consistent with a tumor mass of undetermined origin, but that it might

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be associated with or be a result of congenital deformity. Dr. Ernest Burrows of the Neurological Department then saw the child in consultation; he believed that she was suffering from a developmental anomaly of the spine and cord, but that she was entitled to exploration to rule out the presence of tumor.

Operation. On Jan. 16, 1947, an incision was made centering on the 10th dorsal spine, and the spines and laminae from the 9th to the 12th dorsal vertebrae were

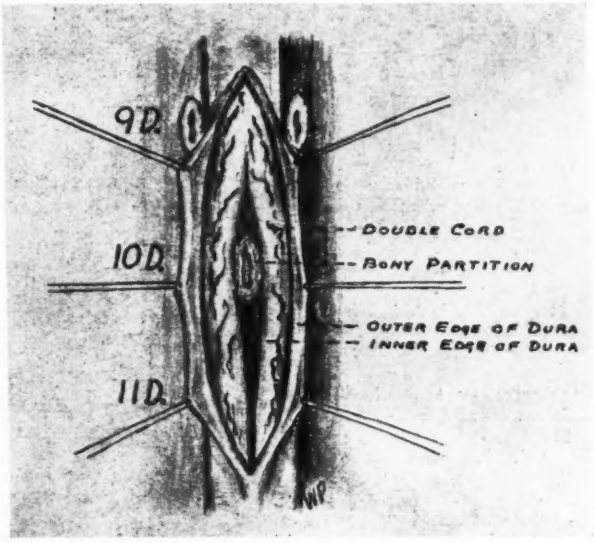

FIG. 2. Drawing of the condition found at operation after removal of the spines and laminae, with the membranes opened.

exposed. The spine of the 9th appeared to be normal; that of the 10th was composed of two fused bulbous masses; and the neural arches of the 11th and 12th were incompletely closed. The spinous process and laminae of the 9th vertebra were removed, and the fused mass representing the spinous process of the 10th was then carefully rongeured off. As this mass was being removed it became evident that there was a ventral projection of bone impinging on the dura and its contents. The incompletely fused arches of the 11th and 12th vertebrae were therefore removed to give better exposure, and the removal of the spine and laminae of the 10th was then completed.

The ventral bony projection, which had at first been thought to be somewhat in the nature of a stalactite, was found to pierce the membranes and the cord and to be firmly attached to the underlying vertebral body. The dura was opened above the bony spur and this exposed an apparently normal cord which divided to pass around the bony partition (Fig. 2). Opening of the dura below the obstruction showed that the split in the cord continued, with obvious separation of the two parts, for at least 2 inches. At this point the parts of the cord came to lie in apposition but did not ap-

pear to fuse into a single cord as far as they could be seen. At the point of its attachment to the spinous process the bony partition was thick, about ½ inch across, but it narrowed down like the keel of a yacht as it approached the vertebral body. It was about ¾ inch in length along the canal. Around this bony partition, the divided cord was enclosed in two separate tubes of dura and arachnoid (Fig. 3); below the obstruction the divided cord had a single dural and arachnoid covering with separate pial envelopes. It was found possible to remove the bony partition without injury

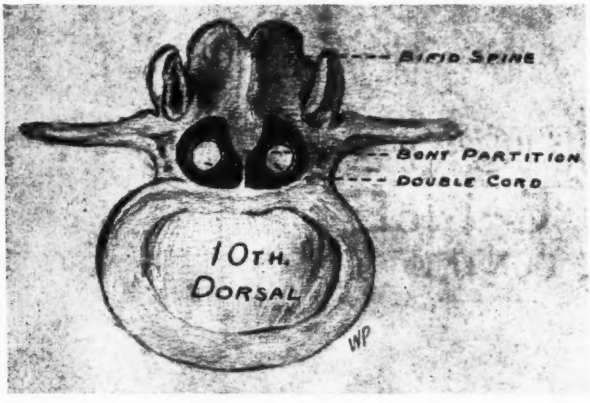

FIG. 3. Cross section of the condition found at operation.

to the nerve elements by rocking it off as one would remove a tooth. The dural envelopes were then retracted and the jagged end of the partition was removed with rongeurs. The dura was closed with interrupted silk stitches and the wound was closed in the usual manner.

Postoperative Course. The girl made a very good recovery. Dr. Burrows was asked to re-examine the child to see if he could discover neurological signs associated with the division of the cord. His findings were essentially the same as those previously recorded and he concluded: "One might expect dissociation of sensation, but there is none. There is probably duplication of the tracts in the cord."

The patient was discharged on her 25th postoperative day. At this time her pain had disappeared, she could move her toes and foot better, and her anesthesia had lessened to a considerable degree.

Addendum

Since the preparation of this paper the girl was examined again on Jan. 13, 1949, 2 years after the operation. She then said that she had no pain and was not troubled by any sense of numbness in the leg. She walked well without any limping. The atrophy of the right leg muscles had disappeared to a large extent, their size and strength being almost equal to that of the corresponding muscles in the other leg. The tendency to inversion of the foot had gone, the height of the arch was less, and the foot itself was almost as large as the one on the left. The knee jerk was still ab

s nt and the ankle jerk was still sluggish. The hypesthesia had disappeared, and the a esthesia of the toes was limited to the three outer ones. Contemplated orthopedic c rrective operation will probably not be needed.

PREVIOUS REPORTS

As neither my associates nor I had ever met a condition similar to this, w were at a loss for an explanation or even for a name until one of my in- t rns, Dr. Thomas Mathieu, came across the article by Marr and Uihlein.[5] This led us to a fairly extensive literature on the subject, which I shall now review briefly.

In an interesting pathological study of the artefacts of the nervous system, Van Gieson,[8] in 1892, described a series of congenital deformities of the cord associated with monstrosities and incompatible with extrauterine life, as follows:

(1) Amyelia—complete absence of the cord associated with absence of the brain.
(2) Atelomyelia—partial development with various sections of the cord missing, usually associated with absence of the brain.
(3) Diastematomyelia—a portion or the whole of the cord split into lateral halves, in which some fusion may occur.
(4) Diplomyelia—true duplication of the cord found in various kinds of double monsters.

Steiner[7] in an inaugural dissertation in 1895 reported 35 cases of cord duplication and added 1 of his own. I have been unable to consult this dissertation, but quote from Bruce, McDonald and Pirie.[1]

In 1906, Bruce, McDonald and Pirie[1] reported a case of partial doubling of the spinal cord. This occurred in a woman 50 years of age who had no evidence of such an abnormality during life, but at postmortem examination was found to show division of the cord in the lumbar region with reunion below.

Zalewska-Ploska,[10] in 1913, reviewed the literature and added 2 cases of her own. The first patient was a 50-year-old woman with spina bifida occulta and hypertrichosis, without symptoms referable to this condition. At postmortem examination she was found to have doubling of the cord at the 2nd lumbar segment. The second patient was an 8-month-old girl with subcutaneous myelomeningocele and doubling of the cord. In very beautiful plates, the author shows that there were two complete cords in the lumbar region, rotated at 90° to each other, each with a central canal and a ventral fissure. In this instance, the condition was found at operation, but the patient did not survive.

Weil and Mathews,[9] in 1935, reported the case of a 5-month-old child, with meningocele and spina bifida, who died of an ascending urinary infection. Postmortem examination showed that the cervical cord was normal. The middorsal region showed hydromyelia, and the lower dorsal cord was

divided completely. Sections showed mirror images turned at an angle of 90 with the anterior fissures facing each other. There were two separate anterio horns in each cord, but the posterior horns were united into one at the middl of the lateral circumference of each cord. There were also elevated cylindrica ependymal cells in each central canal.

In 1936, Hamby[2] reported doubling of the cord in a young woman of 20. In this case, there was a history of spina bifida in the female members o the family. The patient had pain in the lumbosacral area radiating to th right hip and leg, with stiffness of the right leg, and there was a profus growth of hair over a dimple in the lumbar area. The right knee jerk wa slightly exaggerated, there was anesthesia in the saddle area, and a band o hypesthesia down the backs of both legs. X-rays showed distortion of th laminae of the 3rd, 4th, and 5th lumbar vertebrae and most of the sacrum with a failure of fusion in the midline, and congenital fusion of the bodies of the 4th and 5th lumbar vertebrae. At operation, a hair cyst was found underlying the dimple, with a pedicle extending down to a bony spur attached to the vertebral body, the cord being completely separated around this spur. The patient made a good recovery from the operation.

Lichenstein,[4] in 1940, made an interesting and quite exhaustive study of spinal dysraphism, in the course of which he reported 1 instance of cord doubling. In this case there was hydromyelia of the thoracolumbar segments, separation of the central canal into two main branches, and disastasis of the lower cord into two completely separated portions with a common dural envelope. There were secondary diverticulum-like outpouchings of the central canal with multiple atypical central canal formations, and fusion of the subependymal gray substance into irregular areas of so-called primary ependymal gliosis. He felt that this was diastematomyelia and not a true duplication, and he came to the conclusion that all previously reported cases were only pseudoduplications, and that no true doubling had ever been demonstrated.

Later in 1940 Herren and Edwards[3] wrote an excellent review of the reports up to that time. They studied all available records and selected 42 cases as having true duplication of the cord, adding 1 more report of a patient on whom they had operated. The condition was found to be variously named as doubling, duplication, reduplication, bifurcation, and diastematomyelia, but they preferred to call it diplomyelia. It occurred about equally in the 2 sexes, from fetal life to 76 years of age, but only $\frac{1}{4}$ of the cases were found in adolescent or adult life. More than $\frac{1}{2}$ of the instances were associated with spina bifida, usually occult. In practically all cases where there was associated meningocele or myelomeningocele death occurred at an early age, probably from infection. The spinal canal was found to show deformity varying from moderate dilatation to partial doubling; this latter condition was found in of the cases in which there was a midline cone-shaped osseous or osseochondromatous process with its base attached to or continuous with the dorsal aspect of one or several vertebral bodies and with its apex directed into th

lumen of the spinal canal. This process was usually single but there were occasionally two or more; in some instances, a band of connective tissue, sometimes containing cartilage, took the place of the bony spur. Defects in the vertebral bodies, consisting of underdevelopment or absence of several bony segments, occurred in 10 per cent of the cases. The extent of duplication was usually limited to about 10 spinal segments in the lower dorsal or lumbar region, only 2 cases being found above the middorsal area. In all instances the cords were lateral to each other, and each appeared to have rotated through an angle approaching 90°, so that the ventral gray columns of the two cords faced each other and the ventral fissures lay in about the same frontal plane. In several cases, each cord was associated with four primary nerve roots; a well-formed pair of dorsal and ventral roots lying laterally and a rudimentary pair lying medially, these latter roots being usually associated with underdeveloped gray columns. Where doubling occurred, it usually continued down through the sacral segments, but in a few instances there was fusion below, with the re-establishment of a single well-formed cord. There was occasionally hydromyelia in the cord above the division. Abnormal development of the brain was definitely an exception to the usual finding. The most common associated deformity was club foot; there was 1 instance of double ureter and kidneys. No specific clinical features were found to be common to the cases studied.

These authors felt that this condition cannot be explained by arrested development, as there is no normal embryonic stage in which the cord is double. They considered it a primary alteration of the cord, with the bony changes secondary, as the cord is laid down long before the bone. They pointed out that, in the chick embryo, as the lateral extremities of the neural plate approach the midline dorsally they may be directed ventrally for a short distance. Usually they retreat dorsally, fuse, and unite to complete the tube. If they should continue ventrally, they might fuse in such a way as to form two medullary tubes which might go on to form two separate cords. If the distance between these cords were small, the undifferentiated mesenchyme might develop into pia mater; if it were greater, arachnoid, dura, connective tissue, cartilage, or bone could form in this area. After careful study, they came to believe that true doubling of the cord represents an incipient form of twinning.

The patient on whom they operated was a 23-year-old woman with a left club foot which had ulcerated many times. The muscles of this leg were atrophic, and there were no knee or ankle jerks. There was fusion of the bodies from the 8th to the 11th dorsal vertebrae and there were bifid spines from this point to the 3rd lumbar vertebra. At operation, it was found that the arch of the 11th dorsal vertebra was inverted so that it bisected the cord, and was attached to the underlying body. This partition was removed and the patient a made good recovery, to die of another disease about 5 months later. Postmortem examination showed division of the cord from the 7th dorsal level down, with a common dural envelope except at the region of the

bony partition. In the area where the dural tubes were doubled, there was a dentate ligament from the medial aspect of each cord. The pathological findings in the cords were similar to those which they had previously described.

Marr and Uihlein,[5] in 1944, reported operating on a 12-year-old girl with upper thoracic scoliosis and kyphosis, club feet, and spastic gait. The left lower extremity was hypersensitive to cold, and there was numbness of the left great toe. The left leg was weak, the toes on this side could not be dorsiflexed, and there was some incontinence and retention of urine. X-rays showed fusion of the 3rd and 4th ribs posteriorly on the right, a 2nd lumbar hemivertebra, fusion of the bodies of the 3rd, 4th and 5th lumbar vertebrae, lumbar scoliosis and kyphosis, and sacral spina bifida. Pantopaque studies showed a partial block to the passage of oil at the level of the 3rd lumbar vertebra. The protein in the spinal fluid was 30 mg. per cent. At operation a narrow bone spicule was found extending from the body to the arch of the 3rd lumbar vertebra. At this point the cord was completely divided into two parts for a distance of about 4 cm. The doubled cord had a common covering of dura. The bone spicule was removed, and the patient made a good recovery, being discharged in 15 days.

In 1946, Maxwell and Bucy[6] reported a clinical case of diastematomyelia. This occurred in a child of 9 months who had had a dimple in the lumbar region since birth, but who was otherwise apparently normal, with no motor or sphincter disturbances. X-ray examinations showed anomalous development of the lumbar vertebrae, with a calcareous shadow between the 3rd and 4th vertebrae. At operation a bony spur was found extending dorsally from the body of the 3rd lumbar vertebra. The cord was completely separated into two portions at this point and each part had an individual envelope of dura. The spur was removed and the child made a good recovery. Four months later the child was re-examined; walking and talking were then normal, and the only abnormal neurological finding was a suggestive left Babinski response. The authors followed Lichtenstein in feeling that these cases represent a splitting of the cord into two lateral halves, rather than the formation of two relatively complete cords.

SUMMARY

Duplication of the spinal cord, variously called doubling, reduplication, bifurcation, diastematomyelia, and diplomyelia, may occur in varying degrees. There may be an abortive attempt at doubling which results in partial separation. Entire division into parallel structures over a short or relatively long distance may occur, and the resulting cords may be incomplete, or each may contain practically all the elements of a normal spinal cord. They may be separated by pia only, or there may be arachnoid, dura, connective tissue, cartilage, or bone intervening. In the most marked examples there may even be doubling of the lower spinal column. Some observers[4,6] have felt that only pseudoduplication ever occurs; it does not seem possible to agree with this

contention in view of reports showing two practically complete cords, each with its full complement of nerve roots. The most reasonable explanation of the condition seems to be that offered by Herren and Edwards:[3] that it is a primary alteration of the cord, with secondary bone changes, and that it represents an incipient form of twinning.

Available published reports, covering 46 cases, are reviewed. In most of the patients the condition was found at postmortem examination, and in many instances it was not suspected during life. A recent clinical example from the author's own experience is reported in some detail. This account brings the total number of cases to 47. In 6 instances the condition was found at operation, and 5 of these patients survived.

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CASE REPORTS

CYST OF THE FIFTH VENTRICLE TREATED BY INTRAVENTRICULAR DRAINAGE

DOUGLAS MILLER

Department of Neurosurgery, St. Vincent's Hospital, Sydney, Australia

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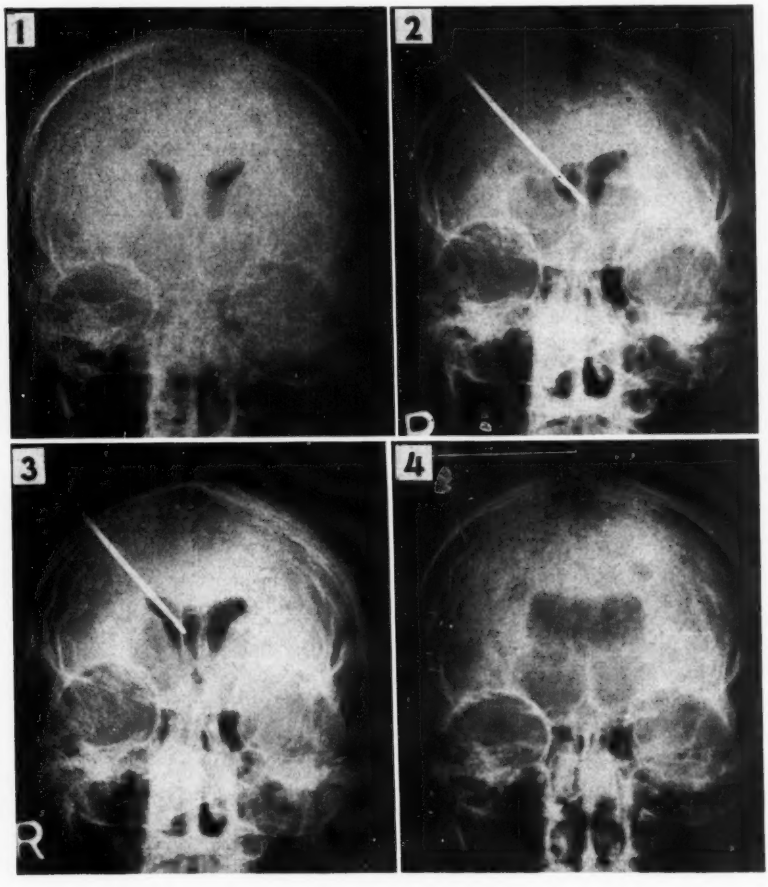
A male, aged 35, while at work became afflicted by a "queer feeling," which was followed by a most intense headache situated at the back of his head and behind the left eye. The



FIGS. 1–4

attack gradually passed off after 3 hours. At intervals of a few days he had similar attacks requiring morphia for their relief. These were succeeded by a constant pain at the back and on the left side of his head. He had no other symptoms.

On examination there was a very early papilloedema of the left fundus. His central nervous system was normal. X-rays of the skull were normal.

In view of the severity and persistence of his symptoms, ventriculography was performed through bilateral occipital burr-holes. The bodies and anterior horns of the lateral ventricles were seen to be separated (Fig. 1).

At a subsequent session, and with radiographic checks during the procedure, a brain cannula was introduced through a right parietal burr-hole, until it was judged to be in the position of the interventricular space. A clear colourless fluid was withdrawn and air injected. The air passed into the left lateral ventricle and filled a cyst of the fifth ventricle. The cannula was withdrawn into the right lateral ventricle which was then filled with air (Figs. 2 and 3).

The following day a radiograph showed air equally distributed in the three cavities. The ventricles showed a general increase in size, characteristic of "second-day encephalograms" (Fig. 4).

It is now over a year since this investigation and the patient has been entirely symptom free.

COMMENT

Cysts of the fifth ventricle are not common. Various rather extensive surgical procedures have been advocated for their treatment. The above case shows that the simple process of establishing an interventricular communication, with a brain cannula, may be effective.

The above procedure could be so readily repeated if symptoms were to recur.

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INTRACRANIAL NEURINOMA OF THE HYPOGLOSSAL NERVE

SUCCESSFUL REMOVAL, CASE REPORT

Michael Scott, M.D., and Henry T. Wycis, M.D.
Department of Neurosurgery, Temple University Hospital and Medical School, Philadelphia, Pennsylvania

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Neurinoma of the 12th cranial nerve is rare.[5,6] Haase[5] reviewed the literature in 1946 and found only 4 cases; 2 of extracranial[2,4] and 2 of intracranial location.[1,3] He added 2 cases of intracranial involvement.

We report the following case:

A white female, aged 50, was referred on July 29, 1945 to the neurosurgical service of one of us (M. S.) by Dr. J. V. Smith of Perth Amboy, N. J.

For 2 years she had had temporal and frontal headaches, which had become severe previous to admission. Numbness and weakness of the right hand had been present for 3 months. Vomiting had occurred almost constantly for 1 month. Prior to admission she began to stagger, hiccup and "to lose her voice." The patient observed that when she ate hard candy (but not other sweetened foods such as coffee, cereal and fruit) she would choke. She insisted that this occurred only when the sweet sensation would reach the back of her throat. She did not have this experience with sour or salty foods and did not remember her reaction to bitter foods. Past medical history was negative except for uncomplicated diphtheria at 36 years of age.

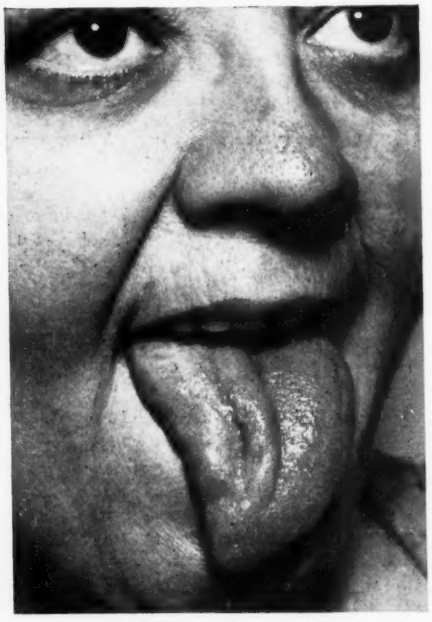

FIG. 1. Paralysis and atrophy of right side of tongue due to neurinoma of hypoglossal nerve.

Neurological Examination. There was fullness of the retinal veins and blurring of the disc margins. There was a decrease in corneal reflex and sensation to pain, temperature, and light touch over the right 1st trigeminal division. The motor portion of the 5th cranial nerve and the 7th and 8th were not impaired. The right soft palate was weak, with absent gag reflex. The right vocal cord was paralyzed. The right trapezius muscle showed slight atrophy and decrease in power. The tongue was completely paralyzed on the right and marked atrophy and fibrillations were present (Fig. 1). The deep tendon reflexes were all active but increased on the left. A bilateral Hoffmann and Oppenheim sign were present. There was no Babinski or Chaddock sign. Except for a weakness in the right hand, motor power was not impaired. No other sensory abnormalities could be detected. Finger-to-nose test revealed marked dysmetria in the right upper limb and bilateral intention tremor, more marked on the right.

Provisional diagnosis: Extramedullary tumor compressing the right ventrolateral surface of the medulla.

Laboratory Findings. Blood count, urinalysis, and serology were negative. Routine stereoscopic X-rays of the skull were negative.

Operation. On July 31, 1945, under endotracheal anesthesia, a high cervical laminectomy and suboccipital craniectomy were done. The posterior rim of the foramen magnum and the first 3 cervical laminae were removed, and the dura was opened by a midline incision. On the right side a large tumor was found in the space between the lateral surface of the medulla and the rim of the foramen magnum. It was encapsulated and firmly wedged in this space (Fig. 2). The tumor had multiple nubbins: one extended mesialward against the medulla; one extended caudad, partly compressing the vertebral artery; another projected cephalad, compressing the inferior surface of the cerebellum. The 9th, 10th, 11th, and 12th cranial nerves were enmeshed in the tumor. The tumor was carefully mobilized from its caudal and mesial aspects. The posterior inferior cerebellar artery was adherent to the upper nubbin and was freed by dissection. It was then seen that the tumor was

connected by a broad base to the jugular and hypoglossal foramina. The base was resected and coagulated. No bone was removed from the hypoglossal or jugular foramen. Following delivery of the tumor, a large space remained in which the following structures were seen: In the caudal portion, the vertebral artery from its place of entrance into the spinal canal to its point of junction with the basilar artery. A portion of the vertebral artery was partially flattened but there was no evidence of any occlusion. The medulla had been completely displaced to the opposite side. The proximal portions of the 9th, 10th, and 11th cranial nerves could be seen but there was no evidence of any part of the 12th nerve, which apparently had been completely enmeshed by the tumor. The dura and the remainder of the incision were then closed in the usual manner.

Microscopic Diagnosis. Neurinoma (Fig. 3).

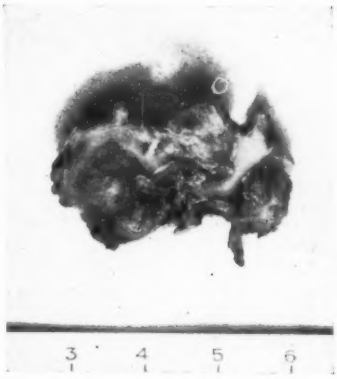

FIG. 2. Tumor removed. Note groove made by vertebral artery.

Postoperative Course. Special right and left posterior oblique stereoscopic projections of the occipital area lateral to the foramen magnum were obtained after operation. The left hypoglossal canal was of normal size and shape. On the right side, in the corresponding region, a large area of bone erosion was noted. The defect involved both the hypoglossal canal and the jugular foramen and measured 2 cm. in diameter (Fig. 4).

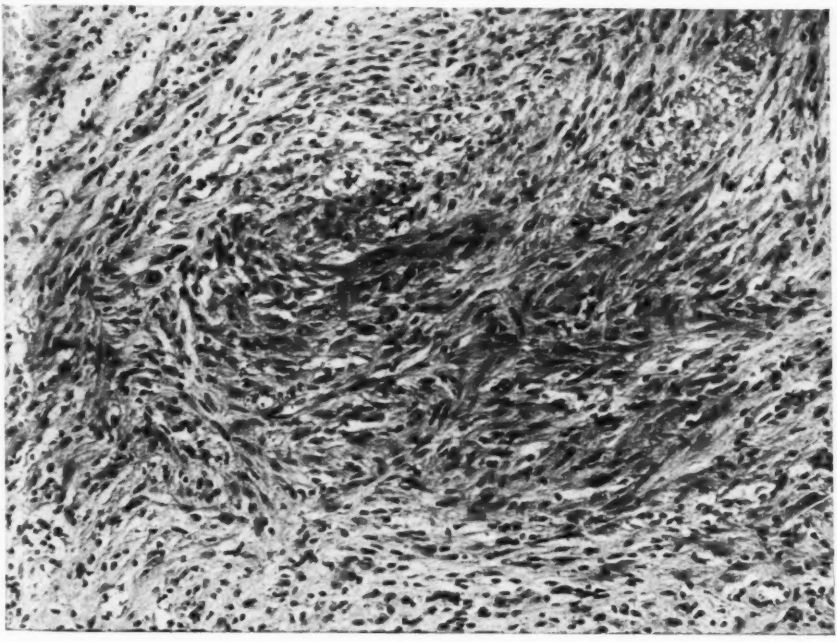

FIG. 3. A section of tumor showing characteristic structure of neurinoma. (Hematoxylin eosin)

FIG. 4. (A) Normal jugular foramen on left side. (B) Large area of bone erosion due to tumor infiltration of jugular and hypoglossal canal.

The patient's convalescence was uneventful and she was discharged 20 days after operation. She was able to swallow liquids and solids well and was free of headaches for the first time in 2 years. She talked in a whisper but her voice was clear. The corneal sensation was acute and the trigeminal nerve showed no involvement. The facial movements were normal and there was no impairment of hearing. The soft palate paralysis, the weakness of the trapezius muscles, and the paralysis of the tongue were unchanged. There was an improvement in power in the right hand and no impairment in the lower extremities. Sensation was normal. A bilateral intention tremor, more marked on the right side, was present. Fine finger movements were slightly impaired on the right. There was a slight swaying in the Romberg position but the tendency to fall to the right had ceased.

The patient was last examined 3 years after operation. She was working regularly and had no complaints. Although the signs of the pre-operative cranial nerve involvement were still present, her voice appeared normal.

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CONCERNING QUECKENSTEDT AND HIS TEST

ARIST STENDER*

Berlin-Charlottenburg, Germany

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IN THE YEAR 1916, Hans Heinrich Georg Queckenstedt published his important article "Zur Diagnose der Rückenmarkskompression" (Dtsch. Z. Nervenheilk., 1916, *55:* 325–333). On the basis of this article the phenomenon described by him has been called Queckenstedt's test. As is well known, it is performed as follows: with the patient lying horizontally on his side a lumbar puncture is made and the cerebrospinal fluid allowed to flow into an attached manometer. If one then compresses the jugular veins in the neck, the level of the fluid in the manometer rises, to fall at once when the pressure is released. If there is, in the spinal canal between the foramen magnum and the lumbar needle, an obstruction that partially or completely occludes the passage, the rise of the fluid following the compression of the veins will be partial or absent. This is called a positive Queckenstedt test.

One must recall that Queckenstedt's discovery was made in that era following the introduction of lumbar puncture by Quincke (1895) in which intensive study of the pathology of the spinal fluid was pursued in all countries, especially in Germany. One learned to know the content of the fluid in cells and protein, especially globulin (Nissl, Nonne-Apelt, Weichbrodt, Pandy). Wassermann discovered his complement-fixation reaction in blood and fluid. A quantitative refinement of the latter made known by Hauptmann led to the introduction of the colloidal reaction by Lange in 1912.

Further the syndrome of compression or obstruction of the fluid by intradural or extradural tumors was described by Froin and Nonne. This phenomenon (increase in protein, usually xanthochromia and no increase in cells) Queckenstedt began to investigate more fully. He was astonished that the chemical and cellular properties of the liquid had been thoroughly investigated while its mechanical behavior had attracted little attention. The increase of the protein content of the fluid without increase of the cells he thought to be oedema of the fluid, corresponding to the stasis oedema of the spinal cord and dura mater, both of which would appear long before a true compression could be spoken of, since any pathological process outside or inside the spinal cord would be accompanied by a swelling (oedema) of the cord and its coverings. Queckenstedt had noted, early in the course of his numerous investigations, that the fluid in the manometer fluctuated with each respiration and that by straining and coughing the pressure immediately moved up and down. In what way he hit upon the maneuver of com-

* Professor extraordinary for Neurology and Neurosurgery in the "Free University" at the West End Hospital, Berlin-Charlottenburg, Germany.

pressing the jugular veins is not evident from the original work, probabl entirely from theoretical physical reasoning, which proceeded from th datum that an increase in the pressure within the cranium—as, for exampl with tumors—would be visible also in the lumbar fluid, provided no hin drance in the spinal canal made the movement of the fluid and, therefore, it communication with the lumbar manometer impossible. Queckenstedt had

HANS HEINRICH GEORG QUECKENSTEDT

with great acuity, remarked that, in the presence of a block of the spina canal, the rise of the column of liquid in the manometer was lacking, whe one compressed the veins, although a rise still occurred by straining an coughing, whereas under normal conditions no significant difference coul be observed between the effect of the straining and of the obstruction of th jugular veins. He concluded correctly from this that a congestion of bloo occurred in the cranium from the obstruction of the cervical veins, tha thus the room for the fluid was reduced, and that thereby it would be dis placed by the resulting increase in pressure which was transmitted caudal

wards with great speed, to become apparent in the manometer. In the presence of a compression of the canal, on the contrary, the rise of the fluid is delayed, incomplete or completely absent, and also following release of the constriction of the neck the pressure sinks slowly or not at all.

It is apparent that the discovery of this phenomenon was of great value for clinical neurology, especially for the diagnosis of processes that encroach upon the spinal canal and impede the circulation of the spinal fluid within the canal.

Doubtless also the discovery of Queckenstedt influenced studies of the physiology of the spinal fluid. This began around the 20's and, especially in the U.S.A., led to new, pioneering knowledge (Cushing, Weed, Dandy) of the secretion and resorption of the fluid under normal and pathological conditions. Modern neurosurgery, developed in the U.S.A., has profited in unforeseen ways and Queckenstedt was thus significantly concerned in its triumphal progress.

Biographical data concerning Queckenstedt's life are scanty. We are indebted for most of them to his nephew, Dr. H. Queckenstedt. He was born in Leipzig-Reudnitz on the fifteenth of August, 1876, the son of a teacher. His ancestors were mostly farmers or teachers. His youth was rather penurious, his father receiving a yearly salary of only 300 "Thalers." Since Queckenstedt, in his early years, showed evidence of superior gifts, every effort was made to enable him to attend high school and university. In school he was especially interested in natural sciences, physics and mathematics. His aptitude for mathematics was such that his brother, who pursued advanced studies in mathematics, whereas Queckenstedt had not, in later years sought his help in the solution of specially difficult mathematical problems. His love of natural science led him while a student, together with friends, to undertake hikes through the woods in order to pursue botanical and zoological studies and—through catching and later selling of vipers—to augment his meagre financial resources.

Two of his best friends are still living—Prof. Hans Curschmann, the internist in Rostock and Prof. H. Bittorf, internist in Leipzig, formerly in Breslau. To these gentlemen we are indebted for many personal and medical details concerning Queckenstedt. They remember him as a very able and enthusiastic physician and investigator, as an absolutely dependable, upright and truthloving character. After finishing his medical studies at the University of Leipzig he passed the state examination in May, 1900, and entered the service of Hofrat Ganser (discoverer of the syndrome bearing his name) in the Dresden Hospital for Mental Diseases, then the Municipal Hospital in Zwickau, where he wrote his Doctoral Dissertation (1904) on "Carcinosarcoma." Later he worked in the medical polyclinic in Heidelberg and went then as assistant to Geheimrat Martius at the medical clinic in Rostock. There he gained the rank of "Privat Dozent" with his work entitled "Researches on the metabolism of iron in pernicious anemia with remarks about the metabolism of iron in general." (Z. klin. Med., 1913,

79: 49–102), and was, in 1918, appointed "extraordinary" professor. Shortl before the end of the war, in which he participated as a consulting in ternist, he was chosen chief physician to the medical section of the cit hospital of Harburg, near Hamburg. Two days before the Armistice he die in the field, being thrown from a shying horse and run over by a heavy muni tions truck. He was not married.

It is true that his scientific production was small in relation to his natura gifts, but we must remember that his scientific work was greatly hindered b war service. Also, Prof. Curschmann says that he never had the itch to write. His chief, Martius, was an outspokenly theoretical worker and left to Queckenstedt in large measure the time-consuming labor of the clinical service. Besides those noted already, he wrote another work on "Changes in the spinal fluid in diseases of the peripheral nerves, especially polyneuritis and sciatica" (Dtsch. Z. Nervenheilk., 1917, *57:* 316–329) and showed for the first time the increased protein observed in such cases. His friends say that he wrote other less well known articles, among these during the war one on "Slight typhoid involvements, especially typhoid periostitis in vaccinated patients." These articles, due to present unfavorable conditions in Germany, we have been unable to locate.

Since interest in investigating the peculiarities of the spinal fluid was in the air, as it were, at the time, it was probably no accident that it was Queckenstedt who discovered his phenomenon since, as we already know, he had a special innate gift for physical and mathematical problems. His test soon became known everywhere as was to be expected. Nowadays it belongs to the armamentarium which a young neurologist has to master in the very first days of his training. Probably no lumbar puncture is done nowadays in a neurological or neurosurgical hospital or clinic without Queckenstedt's test being automatically made. In the meantime, the method has been improved upon. One no longer has to compress the cervical veins brutally by hand but can use an inflatable rubber cuff (Viets) to regulate the compression more delicately. Also one has begun to measure the pressure of the fluid both in the cisterna magna and the spinal sac. With the patient lying down, both suboccipital and lumbar regions are punctured (Ayer, Tönnis) and manometers attached. At intervals of five seconds the pressure of the fluid after compression (Stookey) is read and registered on a curve. If differences in the speed with which the liquor rises, or in the pressure of the fluid, are found, it is possible in good time to demonstrate visually the earliest manifestations of a beginning obstruction to the vertebral canal between the occipital foramen and the lumbar spine.

Queckenstedt died at the age of 42. He was a casualty of the first World War. His gifts, character and training were such as to have led us to expec many other important contributions if he had lived longer.

THE AMERICAN BOARD OF NEUROLOGICAL SURGERY, INC.

COMPLETE LIST OF DIPLOMATES TO JUNE 1, 1949*

(Date appearing to the left of the name indicates the year of certification)

19[illegible] ABBOTT, KENNETH H. 463 East Town St., Columbus 15, Ohio
19[illegible] ABBOTT, WALTER D. 900 Des Moines Bldg., Des Moines 9, Iowa
19[illegible] ADSON, ALFRED W. 102 Second Ave., S.W., Rochester, Minn.
19[illegible] ALEXANDER, EBEN, JR. Bowman Gray School of Medicine, Winston-Salem 7, N. C.
19[illegible] ALLEN, SAMUEL S. University Hospital, Ann Arbor, Mich.
19[illegible] ANDERSON, FRANK M. 1052 West 6th St., Los Angeles 14, Calif.
19[illegible] †ANDERSON, GILBERT C. New Orleans, La.
19[illegible] ARNOLD, JAMES G., JR. The Latrobe, Charles & Read Sts., Baltimore 2, Md.
19[illegible] BACON, ARTHUR H. 203 K.F.H. Bldg., Wichita, Kan.
19[illegible] BAGLEY, CHARLES, JR. The Latrobe, Charles & Read Sts., Baltimore 2, Md.
19[illegible] BAILEY, PERCIVAL 912 South Wood St., Chicago 12, Ill.
19[illegible] BAKER, GEORGE S. 102 Second Ave., S.W., Rochester, Minn.
1949 BALLANTINE, H. THOMAS, JR. 319 Longwood Ave., Boston 15, Mass.
19[illegible] BASSETT, ROBERT C. University Hospital, Ann Arbor, Mich.
1941 BELLOWS, ROWLAND T. 222 Professional Bldg., Charlotte, N. C.
1949 BENNETT, THOMAS S. 388 Young Hotel Bldg., Honolulu 9, Hawaii
1947 BERNSTEIN, LEON L. Winter Veterans Administration Hospital, Topeka, Kan.
1948 BERTRAND, CLAUDE M. 418 Est, Rue Sherbrooke, Montreal, Canada
1943 BESWICK, WILLIAM F. 140 Linwood Ave., Buffalo 9, N. Y.
1947 BLACK, HOWARD A. 2100 Capitol Ave., Sacramento 16, Calif.
1942 BOLDREY, EDWIN B. University of California Medical School, San Francisco 22, Calif.
1941 BRADEN, SPENCER 1342 Hanna Bldg., Cleveland 15, Ohio
1940 BRADFORD, F. KEITH 410 Hermann Professional Bldg., Houston 5, Texas
1943 BRAGDON, FLOYD H. Mercy Hospital, Pittsburgh 19, Pa.
1949 BRIDGERS, WILLIAM H. 111 South Queen St., Columbia, S. C.
1948 BRIDGES, THOMAS J., JR. 1249 Fifth Ave., New York 29, N. Y.
1943 BRODY, BERNARD S. 235 Bishop St., New Haven 11, Conn.
1940 BROWDER, JEFFERSON 200 Hicks St., Brooklyn 2, N. Y.
1941 BROWN, HOWARD A. 384 Post St., San Francisco 8, Calif.
1949 BROWN, M. HUNTER 1033 Gayley Ave., Westwood Village, Los Angeles 24, Calif.
1941 BUCHSTEIN, HAROLD F. 1837 Medical Arts Bldg., Minneapolis 2, Minn.
1944 BUCKLEY, RICHARD C. 50 Farmington Ave., Hartford 5, Conn.
1940 BUCY, PAUL C. 25 East Washington St., Chicago 2, Ill.
1947 BUERMANN, AUGUST 153 Irvington Ave., South Orange, N. J.
1941 BUNTS, ALEXANDER T. 2020 East 93rd St., Cleveland 6, Ohio
1942 CAMPBELL, ELDRIDGE H. 32 Willett St., Albany 6, N. Y.
1947 CAMPBELL, JAMES B. Menninger Foundation, Topeka, Kan.
1948 CARDENAS Y C., JUAN Tabasco 264, Mexico, D. F.
1942 CARMICHAEL, FRANCIS A., JR. 411 Alameda Rd., Kansas City 2, Mo.
1946 CARMODY, JOHN T. B. 340 Main St., Worcester 8, Mass.
1948 CASCINO, JOSEPH P. 25 East Washington St., Chicago 2, Ill.
1949 CHAMBERS, JOHN W. 18 West Franklin St., Baltimore 1, Md.
1946 CHENAULT, HARVEY 200 West Second St., Lexington 15, Ky.
1940 CLEVELAND, DAVID 324 East Wisconsin Ave., Milwaukee 2, Wis.
1941 CLOWARD, RALPH B. 388 Alexander Young Bldg., Honolulu 9, Hawaii
1940 COBURN, DONALD F. 221 Plaza Time Bldg., Kansas City 2, Mo.
1940 COHEN, IRA 4 East 95th St., New York 28, N. Y.

* Diplomates are requested to send any correction in addresses to the Secretary of the Board, Dr. William J. German, New Haven Hospital, New Haven 4, Connecticut.

† Deceased June 27, 1948.

1940 COLEMAN, CLAUDE C. 1200 East Broad St., Richmond 19, Va.
1943 CONE, WILLIAM V. 3801 University St., Montreal 2, Canada
1949 CORRADINI, EVERETT W. 185 East 40th St., Brooklyn 3, N. Y.
1940 CRAIG, WINCHELL McK. 102 Second Ave., S.W., Rochester, Minn.
1943 CRAMER, FRITZ 700 West 168th St., New York 32, N. Y.
1940 CRAWFORD, ALBERT S. Henry Ford Hospital, Detroit 2, Mich.
1947 CROCKETT, HERBERT G. 511 South Bonnie Brae St., Los Angeles 5, Calif.
1940 CRUTCHFIELD, W. GAYLE University of Virginia Hospital, Charlottesville, Va.
1947 CUNEO, HENRY M. 523 West 6th St., Los Angeles 14, Calif.
1940 *DANDY, WALTER E. Baltimore, Md.
1949 DANIELS, JAMES T. 1028 Park Ave., New York 28, N. Y.
1940 DAVIDOFF, LEO M. 815 Park Ave., New York 21, N. Y.
1947 DAVIS, EDWARD W. 806 S.W. Broadway, Portland 5, Ore.
1940 DAVIS, LOYAL 700 North Michigan Ave., Chicago 11, Ill.
1948 DECKER, HENRY G. 503 Equitable Bldg., Des Moines 9, Iowa
1941 DEERY, EDWIN M. 563 Park Ave., New York 21, N. Y.
1946 D'ERRICO, ALBERT 1430 Medical Arts Bldg., Dallas 1, Texas
1945 DONAGHY, R. M. PEARDON Mary Fletcher Hospital, Burlington, Vt.
1946 DORSEY, JOSEPH F. 270 Commonwealth Ave., Boston 16, Mass.
1946 DOWMAN, CHARLES E. 1028 West Peachtree St., N.W., Atlanta 6, Ga.
1948 EBIN, JUDAH, 935 Park Ave., New York 28, N. Y.
1942 ECHLIN, FRANCIS A. 555 Park Ave., New York 21, N. Y.
1941 ECHOLS, DEAN H. 3503 Prytania St., New Orleans 15, La.
1941 ECKER, ARTHUR 608 East Genesee St., Syracuse 2, N. Y.
1946 EHNI, GEORGE Scott and White Clinic, Temple, Texas
1940 EHRLICH, WILLIAM 31 Lincoln Park, Newark, N. J.
1949 EISENBEISS, JOHN A. 926 E. McDowell Road, Phoenix, Ariz.
1949 ELKINS, CHARLES W. 820 Hanna Bldg., Cleveland 15, Ohio
1947 ELVIDGE, ARTHUR R. 3801 University St., Montreal 2, Canada
1942 ERICKSON, THEODORE C. Wisconsin General Hospital, Madison 6, Wis.
1947 ETTLESON, ABRAHAM 1052 West 6th St., Los Angeles 14, Calif.
1940 EVANS, JOSEPH P. Cincinnati General Hospital, Cincinnati 29, Ohio
1940 FAY, TEMPLE 8811 Germantown Ave., Philadelphia 18, Pa.
1947 FEIRING, EMANUEL H. 815 Park Ave., New York 21, N. Y.
1943 FENDER, FREDERICK A. Stanford University Hospital, San Francisco 15, Calif.
1940 FINCHER, EDGAR F. 384 Peachtree St., N.E., Atlanta 3, Ga.
1947 FINLAYSON, A. I. 1527 Medical Arts Bldg., Omaha, Neb.
1941 FITCH, THOMAS S. P. 916 Park Ave., Plainfield, N. J.
1940 FLEMING, HOWARD W. 384 Post St., San Francisco 8, Calif.
1946 FREED, CHARLES G. 550 Metropolitan Bldg., Denver 2, Colo.
1947 FRENCH, J. DOUGLAS Birmingham Veterans Administration Hospital, Van Nuys, Calif.
1948 FRENCH, LYLE A. University Hospital, Minneapolis 14, Minn.
1947 FULCHER, O. HUGH 1150 Connecticut Ave., N.W., Washington 6, D. C.
1940 FURLOW, LEONARD T. 530 Beaumont Medical Bldg., St. Louis 8, Mo.
1948 GAGE, EVERETT L. Bluefield Sanitarium, Bluefield, W. Va.
1946 GALBRAITH, JAMES G. 1117 South 22nd St., Birmingham, Ala.
1948 GALLAGHER, JOHN P. 2023 R. St., N.W., Washington 9, D. C.
1940 GARDNER, W. JAMES 2020 East 93rd St., Cleveland 6, Ohio
1947 GAROL, HUGH W. 634 East Santa Clara St., San Jose 12, Calif.
1940 GERMAN, WILLIAM J. New Haven Hospital, New Haven 4, Conn.
1941 GLASS, ROBERT L. 608 Hume Mansur Bldg., Indianapolis 4, Ind.
1941 GOTTEN, NICHOLAS 22 North Manassas, Memphis, Tenn.
1949 GOVONS, SIDNEY R. 803 Cathedral St., Baltimore, Md.
1948 GRAF, CARL J. 307 Linwood Ave., Buffalo 9, N. Y.
1940 GRANT, FRANCIS C. 3400 Spruce St., Philadelphia 4, Pa.
1942 GRANT, WILLIAM T. 1136 West 6th St., Los Angeles 14, Calif.
1942 GRANTHAM, EVERETT G. 405 Heyburn Bldg., Louisville 2, Ky.
1948 GREEN, JOHN R. 910 Professional Bldg., Phoenix, Ariz.

* Deceased April 19, 1946.

1941 GREENWOOD, JAMES, JR. 1105 Hermann Professional Bldg., Houston 5, Texas
1941 GRINO, ANTONIO St. Vincent's Hospital, New York 11, N. Y.
1941 GROFF, ROBERT A. 1930 Chestnut St., Philadelphia 3, Pa.
1941 GROSS, SIDNEY W. 8 East 83rd St., New York 28, N. Y.
1941 GRUNNAGLE, JEROME F. 516 Medical Arts Bldg., Pittsburgh 13, Pa.
1941 GURDJIAN, ELISHA S. 840 David Whitney Bldg., Detroit 26, Mich.
1941 GUSTAFSON, WESLEY A. 700 North Michigan Ave., Chicago 11, Ill.
1941 DE GUTIÉRREZ-MAHONEY, C. G. St. Vincent's Hospital, New York 11, N. Y.
1941 HAMBY, WALLACE B. 140 Linwood Ave., Buffalo 9, N. Y.
1941 HAMLIN, HANNIBAL 4 George St., Providence 6, R. I.
1941 HASTINGS, WARREN C. 811 Ewing St., Fort Wayne 2, Ind.
1941 HAVEN, HALE 1115 Terry Ave., Seattle 1, Wash.
1941 HAVERFIELD, W. TRACY 1001 DuPont Bldg., Miami 32, Fla.
1941 HAWKES, C. D. 22 North Manassas, Memphis, Tenn.
1941 HAYNE, ROBERT A. Springer Clinic, 604 South Cincinnati Ave., Tulsa, Okla.
1941 HERRMANN, JESS D. 521 N.W. 11th, Oklahoma City 3, Okla.
1941 HEUSNER, A. PRICE Boston City Hospital, Boston 18, Mass.
1941 HEYL, HENRY L. Hitchcock Clinic, Hanover, N. H.
1941 HOEN, THOMAS I. 1249 Fifth Ave., New York 29, N. Y.
1941 HOLBROOK, THOMAS J. First Huntington National Bank Bldg., Huntington 1, W. Va.
1941 HOLLISTER, NATHANIEL R. 2676 Salem Ave., Dayton 6, Ohio
1941 HORRAX, GILBERT 605 Commonwealth Ave., Boston 15, Mass.
1941 HURTEAU, EVERETT F. 175 South Main St., Akron 8, Ohio
1941 HYNDMAN, OLAN R. 621 First National Bldg., Davenport, Iowa
1941 INGRAHAM, FRANC D. 300 Longwood Ave., Boston 15, Mass.
1942 JAEGER, J. RUDOLPH Jefferson Medical College, Philadelphia 2, Pa.
1942 JELSMA, FRANKLIN 421 Heyburn Bldg., Louisville 2, Ky.
1941 JOHNSON, HERBERT C. Johns Hopkins Hospital, Baltimore 5, Md.
1944 JONES, DEWARD W. 910 Professional Bldg., Los Angeles 14, Calif.
1940 JONES, OTTIWELL W., JR. 384 Post St., San Francisco 8, Calif.
1948 JONES, W. A. Veterans Administration Hospital, Los Angeles 25, Calif.
1940 KAHN, EDGAR A. University Hospital, Ann Arbor, Mich.
1947 KANE, J. WORDEN 55 Riverside Drive, Binghamton, N. Y.
1942 KAPLAN, ABRAHAM 30 East 40th St., New York 16, N. Y.
1946 KARR, HOWARD H. 1521 Delachaise St., New Orleans 15, La.
1940 KEEGAN, J. JAY 1527 Medical Arts Bldg., Omaha, Neb.
1948 KERN, CARROLL E. Veterans Administration Hospital, McKinney, Texas
1940 KLEMME, ROLAND M. 4952 Maryland Ave., St. Louis 8, Mo.
1948 KLEMPERER, WOLFGANG W. 501 Medical-Dental Bldg., Seattle 1, Wash.
1948 KLOOS, EDWARD K. 919 Taylor St. Bldg., Portland 5, Ore.
1948 KOSKOFF, YALE D. 201 South Craig St., Pittsburgh 13, Pa.
1947 KRUEGER, ERICH G. 633 Riverside Drive, New York 31, N. Y.
1947 LAMBROS, VASILIOS S. 7720-14th St., N.W., Washington, D. C.
1941 LAWRENCE, LESTER B. 411 Thirtieth St., Oakland 9, Calif.
1943 LEFEVER, HARRY E. 9 Buttles Ave., Columbus 8, Ohio
1947 LEHMANN, PETER 736 Granville St., Vancouver, B. C.
1941 LIGHT, RICHARD U. 828 West South St., Kalamazoo, Mich.
1941 LIPSCOMB, WILLIAM R. 1809 East 18th Ave., Denver 6, Colo.
1941 LIST, CARL F. Medical Arts Bldg., 26 Sheldon Ave., Grand Rapids 2, Mich.
1948 LIVINGSTON, KENNETH E. 806 S.W. Broadway, Portland 5, Ore.
1947 LOTSPEICH, EDGAR S. 2314 Auburn Ave., Cincinnati 19, Ohio
1940 LOVE, J. GRAFTON 102 Second Ave., S.W., Rochester, Minn.
1944 LOWIS, SAMUEL 475 Commonwealth Ave., Boston 15, Mass.
1944 LUSIGNAN, FRANK W. 450 Sutter St., San Francisco 8, Calif.
1940 LYERLY, JAMES G. 516 Greenleaf Bldg., Jacksonville 2, Fla.
1947 MACCARTY, COLLIN S. 102 Second Ave., S.W., Rochester, Minn.
1941 MCCRAVEY, AUGUSTUS 410 Medical Arts Bldg., Chattanooga, Tenn.
1947 MACKAY, HUNTER J. 1320 Madison St., Seattle 4, Wash.
1943 MCKENZIE, KENNETH G. 430 Medical Arts Bldg., Toronto 5, Canada
1941 MCKINNEY, WILLIAM W. Medical Arts Bldg., Fort Worth 2, Texas

1949 MacLean, J. Arthur 325 East 57th St., New York 22, N. Y.
1949 McNerney, John C. U. S. Naval Hospital, Bethesda 14, Md.
1949 Mabon, Robert F. 31 North State St., Chicago 2, Ill.
1942 Maltby, George L. 203 State St., Portland 3, Me.
1942 Martin, John 700 North Michigan Ave., Chicago 11, Ill.
1947 Martinez-Galindo, Juan de D. University of Virginia Hospital, Charlottesville, Va.
1940 *Masson, Clement B. New York, N. Y.
1948 Matson, Donald D. 300 Longwood Ave., Boston 15, Mass.
1947 Maxwell, Harry P. 208 East Wisconsin Ave., Milwaukee 2, Wis.
1940 Mayfield, Frank H. 2314 Auburn Ave., Cincinnati 19, Ohio
1948 Meacham, William F. 2122 West End Ave., Nashville 5, Tenn.
1948 Meirowsky, Arnold M. Kennedy Hospital, Memphis 15, Tenn.
1941 Meredith, John M. 1200 East Broad St., Richmond 19, Va.
1942 Meyers, Russell Iowa State University Hospital, Iowa City, Iowa
1944 Michelsen, Jost J. 311 Beacon St., Boston 16, Mass.
1940 Mixter, W. Jason 319 Longwood Ave., Boston 15, Mass.
1940 Morrissey, Edmund J. 450 Sutter St., San Francisco 8, Calif.
1941 Mount, Lester A. 700 West 168th St., New York 32, N. Y.
1946 Mufson, Joseph A. 425 East Wisconsin Ave., Milwaukee 2, Wis.
1940 Munro, Donald 818 Harrison Ave., Boston 18, Mass.
1948 Munslow, Ralph A. 1233 Nix Professional Bldg., San Antonio 5, Texas
1940 Murphey, Francis 899 Madison Ave., Memphis 3, Tenn.
1949 Murphy, J. Peter 1911 R St., N.W., Washington 9, D. C.
1940 Naffziger, Howard C. 384 Post St., San Francisco 8, Calif.
1949 Negrin, Juan, Jr. 1010 Fifth Ave., New York 28, N. Y.
1946 Nielsen, Aage 10 Peterboro St., Detroit 1, Mich.
1941 Norcross, Nathan C. 400-29th St., Oakland 9, Calif.
1944 Nosik, Wiliam A. 10515 Carnegie Ave., Cleveland 6, Ohio
1946 Odom, Guy L. Duke University, Durham, N. C.
1940 Oldberg, Fric 912 South Wood St., Chicago 12, Ill.
1944 Oljenick, Ignatius W. 435 East 57th St., New York, N. Y.
1944 Olsen, Axel Guthrie Clinic, Sayre, Pa.
1945 Otenasek, Frank J. 6 East Eager St., Baltimore 2, Md.
1949 Parsons, Herbert 57 East 93rd St., New York 28, N. Y.
1947 Patton, William B. 2205 Highland Ave., Birmingham, Ala.
1940 †Peet, Max M. Ann Arbor, Mich.
1943 Penfield, Wilder 3801 University St., Montreal 2, Canada
1948 Perret, George Iowa State University Hospital, Iowa City, Iowa
1940 Peyton, William T. University of Minnesota Hospital, Minneapolis 14, Minn.
1940 Pilcher, Cobb Vanderbilt University Hospital, Nashville 4, Tenn.
1947 Polmeteer, Frank E. 716 Professional Bldg., Long Beach 2, Calif.
1942 Pool, J. Lawrence 710 West 168th St., New York 32, N. Y.
1943 Poppen, James L. 605 Commonwealth Ave., Boston 15, Mass.
1948 Pribram, Karl H. 333 Cedar St., New Haven 11, Conn.
1943 Pudenz, Robert H. 696 East Colorado St., Pasadena 1, Calif.
1940 Putnam, Tracy J. 416 North Bedford Drive, Beverly Hills, Calif.
1947 Quade, Raymond H. 104 North Commercial St., Neenah, Wis.
1941 Raaf, John 912 Medical-Dental Bldg., Portland 5, Ore.
1940 Rand, Carl W. 523 West 6th St., Los Angeles 14, Calif.
1945 Raney, Aidan A. 1136 West 6th St., Los Angeles 14, Calif.
1940 Raney, Rupert B. 1136 West 6th St., Los Angeles 14, Calif.
1947 Raskind, Robert 899 Madison Ave., Memphis 3, Tenn.
1943 Rasmussen, Theodore University of Chicago, Chicago 37, Ill.
1942 Ray, Bronson S. 525 East 68th St., New York 21, N. Y.
1940 Reeves, David L. 316 West Junipero St., Santa Barbara, Calif.
1941 Reichert, Frederick Leet Stanford University Hospital, San Francisco 15, Calif.
1946 Risteen, Wilford A. University Hospital, Augusta, Ga.
1948 Rizzoli, Hugo V. 1150 Connecticut Ave., Washington 6, D. C.

* Deceased September 5, 1946.
† Deceased March 25, 1949.

194 ROBERTSON, ROBERT C. L. 411 Hermann Professional Bldg., Houston 5, Texas
194 ROULHAC, GEORGE E. Washington University School of Medicine, St. Louis 10, Mo.
194 ROWE, STUART N. 516 Medical Arts Bldg., Pittsburgh 13, Pa.
194 SACHS, ERNEST 333 Cedar St., New Haven 11, Conn.
194 Sanchez-G., Manuel Marsella #14, Mexico, D. F.
194 SCARFF, JOHN E. 710 West 168th St., New York 32, N. Y.
194 SCHLESINGER, BENNO 1125 Madison Ave., New York 28, N. Y.
194 SCHLESINGER, EDWARD B. 710 West 168th St., New York 32, N. Y.
194 SCHNITKER, MAX T. 1031 Secor Hotel Bldg., Toledo 4, Ohio
194 SCHREIBER, FREDERIC 10 Peterboro St., Detroit 1, Mich.
194 SCHULTZ, E. C. 525 Physicians and Surgeons Bldg., Memphis 3, Tenn.
194 SCHWARTZ, HENRY G. Washington University School of Medicine, St. Louis 10, Mo.
194 SCOTT, MICHAEL Temple University Hospital, Philadelphia 40, Pa.
194 SCOVILLE, WILLIAM B. 85 Jefferson St., Hartford 6, Conn.
194 SEGERBERG, LUDWIG H. 405 Heyburn Bldg., Louisville 2, Ky.
194 SEMMES, R. EUSTACE 899 Madison Ave., Memphis 3, Tenn.
194 SHANNON, EDWARD W. 1314 Hanna Bldg., Cleveland 15, Ohio
194 SHELDEN, C. HUNTER 696 East Colorado St., Pasadena 1, Calif.
194 SHENKIN, HENRY A. 1930 Chestnut St., Philadelphia 3, Pa.
194 SHERMAN, IRVING J. 1026 Park Ave., Bridgeport 4, Conn.
194 SIRIS, JOSEPH H. 555 Prospect Place, Brooklyn 16, N. Y.
194 SMOLIK, EDMUND A. 3720 Washington Ave., St. Louis 8, Mo.
194 SNODGRASS, SAMUEL R. John Sealy Hospital, Galveston, Texas
194 SPEIGEL, I. JOSHUA 55 East Washington St., Chicago 2, Ill.
194 SPENCE, WILLIAM T. 1150 Connecticut Ave., Washington 6, D. C.
194 SPURLING, R. GLEN 405 Heyburn Bldg., Louisville 2, Ky.
194 STAFFORD, DONALD E. The Neurosurgical Clinic, 902 Boren Ave., Seattle 4, Wash.
194 STEVENSON, WILLIAM D. 141 Coburg Rd., Halifax, Nova Scotia
194 STOOKEY, BYRON 710 West 168th St., New York 32, N. Y.
194 STOWELL, AVERILL Springer Clinic, 604 South Cincinnati Ave., Tulsa, Okla.
194 STUCK, RALPH M. 632 Republic Bldg., Denver 2, Colo.
194 SUCKLE, HENRY M. 1300 University Ave., Madison 5, Wis.
194 SWANSON, HOMER S. 384 Peachtree St., N.E., Atlanta 3, Ga.
194 SWEET, WILLIAM H. Massachusetts General Hospital, Boston 14, Mass.
194 TAEFFNER, JOHN H. 3464 West School House Lane, Philadelphia 44, Pa.
194 TAFFEL, MAX New Haven Hospital, New Haven 4, Conn.
194 TARLOV, I. M. 1150 Fifth Ave., New York 29, N. Y.
194 TEACHENOR, FRANK R. 221 Plaza Time Bldg., Kansas City 2, Mo.
194 TEXTOR, CHARLES S., II 122 South Michigan Ave., Chicago 3, Ill.
194 THOMPSON, RAYMOND K. The Latrobe, Charles & Read Sts., Baltimore 2, Md.
194 THOMSON, JAMES L. 405 Wainwright Bldg., Norfolk 10, Va.
194 TINSLEY, MILTON 25 East Washington St., Chicago 2, Ill.
194 TITRUD, LEONARD A. 1141 Medical Arts Bldg., Minneapolis 2, Minn.
194 TROLAND, CHARLES E. 1200 East Broad St., Richmond 19, Va.
194 TURNBULL, FRANK 913 Medical-Dental Bldg., Vancouver, B. C., Canada
194 TURNER, OSCAR A. 2204 Glenwood Ave., Youngstown 2, Ohio
194 TURNEY, M. FRANK Medical Arts Bldg., Knoxville, Tenn.
194 UIHLEIN, ALFRED 102 Second Ave., S.W., Rochester, Minn.
194 ULMER, JACK L. Veterans Administration Hospital, Richmond 19, Va.
194 VAN WAGENEN, WILLIAM P. 260 Crittenden Blvd., Rochester 7, N. Y.
194 VER BRUGGHEN, ADRIEN 122 South Michigan Ave., Chicago 3, Ill.
194 VOGEL, PHILIP J. 1801 New Jersey St., Los Angeles 33, Calif.
194 VORIS, HAROLD C. 30 North Michigan Ave., Chicago 2, Ill.
194 WALKER, A. EARL Johns Hopkins Hospital, Baltimore 5, Md.
194 WALKER, EXUM 490 Peachtree St., N.E., Altlanta 3, Ga.
194 WARD, ARTHUR A., JR. University of Washington School of Medicine, Seattle 5, Wash.
194 WATSON, C. ROBERT 1014 Donaghey Bldg., Little Rock, Ark.
194 WATTS, JAMES W. 1911 R. St., N.W., Washington 9, D. C.
194 WEARY, WILLARD B. 4319 Oak Lawn Ave., Dallas 4, Texas
194 WEAVER, THOMAS A., JR. 521 Third National Bldg., Dayton 2, Ohio

1945 WEBSTER, JOHN E. 840 David Whitney Bldg., Detroit 26, Mich.
1941 WEGNER, WALTER R. 818 Harrison Ave., Boston 18, Mass.
1942 WEINBERGER, LAURENCE M. 908 Second National Bldg., Akron, Ohio
1944 WERDEN, DELBERT H. 501 Medico-Dental Bldg., San Diego 1, Calif.
1945 WHITCOMB, BENJAMIN B. 85 Jefferson St., Hartford 6, Conn.
1942 WHITE, JAMES C. Massachusetts General Hospital, Boston 14, Mass.
1946 WHITFIELD, ROBERT D. 38 Willett St., Albany 6, N. Y.
1948 WIGDERSON, HENRY 41-08 74th St., Jackson Heights, L. I., N. Y.
1941 WILKINS, HARRY 521 N.W. 11th, Oklahoma City 3, Okla.
1948 WILLIAMSON, WILLIAM P. 221 Plaza Time Bldg., Kansas City 2, Mo.
1947 WILSON, ARCHER A. 1031 Quarrier St., Charleston 1, W. Va.
1941 WINGEBACH, WILFRID 1045 Park Ave., New York 28, N. Y.
1948 WOLKIN, JULIUS 2010 East 102nd St., Cleveland, Ohio
1943 WOODHALL, BARNES Duke University Hospital, Durham, N. C.
1943 WOODS, WARD W. Medico-Dental Bldg., San Diego 1, Calif.
1947 WOOLF, JACK I. Medical Arts Bldg., Dallas 1, Texas
1949 WOOLSEY, ROBERT D. 16 Hampton Village Plaza, St. Louis, Mo.
1949 WRIGHT, STEWART A. 310 Medical Arts Bldg., Salt Lake City, Utah
1945 WYCIS, HENRY T. Temple University Hospital, Philadelphia 40, Pa.

Summary

Total certified=302. Deceased=4. Living=298.

GEOGRAPHICAL LOCATION OF NEUROSURGEONS CERTIFIED BY THE AMERICAN BOARD OF NEUROLOGICAL SURGERY, INC.

UNITED STATES

ALABAMA (2)

IRMINGHAM
Galbraith, J. G.
Patton, W. B.

ARIZONA (2)

HOENIX
Eisenbeiss, J. A.
Green, J. R.

ARKANSAS (1)

ITTLE ROCK
Watson, C. R.

CALIFORNIA (33)

EVERLY HILLS
Putnam, T. J.
ONG BEACH
Polmeteer, F. E.
OS ANGELES
Anderson, F. M.
Brown, M. H.
Crockett, H. G.
Cuneo, H. M.
Ettleson, A.
Grant, W. T.
Jones, D. W.
Jones, W. A.
Rand, C. W.
Raney, A. A.
Raney, R. B.
Vogel, P. J.
OAKLAND
Lawrence, L. B.
Norcross, N. C.
PASADENA
Pudenz, R. H.
Shelden, C. H.
SACRAMENTO
Black, H. A.
SAN DIEGO
Werden, D. H.
Woods, W. W.
SAN FRANCISCO
Boldrey, E. B.
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Michelsen, J. J.
Mixter, W. J.
Munro, D.
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Sweet, W. H.
Wegner, W. R.
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Schlesinger, E. B.
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Olsen, A.

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Lehmann, P.
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MEXICO, D. F.
Cardenas Y C., J.
Sanchez-G., M.

NOTICES

AMERICAN NEUROLOGICAL ASSOCIATION

At the seventy-fourth annual meeting of the American Neurological Association held in Atlantic City, New Jersey, from June 13–15, 1949, the following officers were elected for the year 1949–1950: President, Dr. Henry W. Woltman; 1st Vice-President, Dr. Johannes M. Nielsen; 2nd Vice-President, Dr. E. Jefferson Browder; Secretary-Treasurer, Dr. H. Houston Merritt; Assistant Secretary, Dr. Charles Rupp.

HARVEY CUSHING SOCIETY

The 17th annual meeting of the Harvey Cushing Society was held in the Yale School of Medicine, New Haven, Connecticut, on June 2, 3 and 4, 1949 in commemoration of Harvey Cushing's eightieth birthday. Over 260 members and guests from the United States, Canada and abroad were present. Scientific sessions were held for three days. Dr. Winchell McK. Craig, Mayo Clinic, Rochester, Minnesota, was the presiding officer. At the annual dinner Dr. Daniel C. Elkin, Emory University, Atlanta, Georgia, gave a superb account of Dr. Cushing in his address "Remembrance of Things Past," which was followed by a series of photographs of "The Chief" from the age of sixty to sixty-six, shown by Dr. Richard U. Light of Kalamazoo, Michigan.

Officers elected for 1949–1950 are: President, Dr. Frank Turnbull, Vancouver, B.C.; Vice-President, Dr. Frank H. Mayfield, Cincinnati; Secretary-Treasurer, Dr. Louise Eisenhardt, New Haven. Members of the Executive Committee are: Dr. Edgar F. Fincher, Atlanta; Dr. Franc D. Ingraham, Boston; Dr. Frank R. Teachenor, Kansas City; Dr. Barnes Woodhall, Durham, and Dr. W. McK. Craig, Rochester.

SCANDINAVIAN NEUROSURGICAL SOCIETY

The 5th meeting of the Scandinavian Neurosurgical Society will take place in Arhus, Denmark, September 14 and 15, 1949. Members of American and other European neurosurgical societies are cordially invited to attend. Information is obtainable from the Secretary, Dr. Olof Sjöqvist, Södersjukhuset, Stockholm, Sweden, or from the host of the meeting, Dr. Richard Malmros, Kommunehospitalet, Arhus.

NATIONAL COMMITTEE FOR CHILE

The Committee is now receiving gifts for the Library of the Medical School of the University of Chile at its new collection center in the Library of Congress, Washington. The newer materials in the library in Chile, including periodicals, books and reference materials, were totally destroyed in the recent fire. Medical periodicals of the last ten years and recent medical books are urgently needed. Contributions will be appreciated, and should be sent to National Committee for Chile, Room 313, Library of Congress, Washington, D. C.

THE SENSORY PATHWAYS FROM THE SHOULDER JOINT

MARTIN WRETE, M.D.
Histological Department, Uppsala University, Uppsala, Sweden

(Received for publication September 21, 1948)

THE anatomy of the peripheral nervous system in man has been engaging increasing attention owing to the development of neurosurgery during the last few decades. One of the reasons why I have taken up the question of the innervation of the shoulder joint for closer examination is the knowledge that a Swedish surgeon, Dr. H. Wahren, in the course of the last few years, has made operative cuts of nerve branches running to the joints, as a last resort in certain states of severe pain. That surgical operations on the nerves of the joints require a thorough knowledge of the anatomy of those nerves is self-evident. From other points of view also, such knowledge is desirable. Pains in the shoulder joint and arms involve many still rather obscure problems and, even if these problems lie mainly in the sphere of physiology and pathology, it seems essential that the normal neuromorphological conditions should be ascertained so far as possible.

If we turn to standard textbooks on anatomy, we find there merely quite summary information on the origin and distribution of the articular nerves, and the same is true of even the large monographs on the peripheral nervous system by Hovelacque[11] and Clara.[4] The older textbooks, such as those by Cruveilhier,[5] Henle,[9] Quain,[20] and Poirier and Charpy,[19] give somewhat fuller accounts. The most detailed descriptions and illustrations of the articular nerves, however, have been given by Rüdinger,[22] who, by minute dissections, followed the nerve branches to the joints as far as seems to have been possible. Unfortunately, however, he does not inform us on how large a material his description is based. A schematic representation of the innervation of the extremity joints, evidently based on Rüdinger's account, can be found in an atlas of the peripheral nerves by Hasse.[8]

By numerous histologic investigations it has been shown that nerve branches run to the articular capsules and their ligaments, to the articular menisci and disci and to the glenoidal labra. That the hyaline cartilages are devoid of nerves is a generally accepted view, which tallies with the observation made by Bloch,[1] Lennander,[14,15] and Nyström[18] that hyaline cartilage is non-sensitive.

As regards the structure of the articular nerves, Rüdinger[22] as far back as 1857 had suggested that the fibres of which they are constituted are of both encephalospinal and sympathetic origin. A more systematic investigation was made by Sherrington[27] on cats and monkeys, and by Sasaoka[23,24] on cats. The principal facts brought to light by these investigations are that the medullated fibres in the articular nerves have a calibre similar to that of the slenderer nerve branches to the skin, and that the articular nerves also

contain non-medullated fibres in the same proportion as in the said nerve branches. These observations thus afford morphological evidence that the joints receive both sensory and sympathetic nerve fibres, as is the case with the skin. The terminal branches of the articular nerves, their sensory nerve endings and fibres to the blood vessels have been described in detail by a large number of authors, such as Nicoladoni,[17] Krause,[13] Sfameni,[26] Gerneck,[7] and Gardner.[6]

The information I have found in the literature regarding the nerves of the shoulder joint in particular is derived almost entirely from Rüdinger's detailed macroscopic description; I have also consulted a minor anatomical investigation by Rauber.[21] These investigations will be more fully reported in connection with the description of my own results.

MATERIAL. TECHNIQUE. TERMINOLOGY

It seemed unlikely that with the usual method, *viz.* dissection with or without the aid of a magnifying-glass, one could contribute appreciably more to the study of the joints than previous investigators, some of whom apparently attained the utmost skill in this technique. It was evident that recourse to other methods would be necessary.

In previous studies of the anatomy of the vegetative nervous system, I had adopted the following procedure: in serial sections of Ag-impregnated human fetuses I followed the nerves in the microscope and afterwards reconstructed them.[30,31,32,33,34] This procedure is laborious, firstly in producing the serial sections and secondly in the execution of the reconstructions; on the other hand, it affords good prospects of making further progress than with the dissection method formerly adopted. One need not, as in dissection, reckon with the possibility that a nerve branch may be overlooked owing to its being torn off before it is detected. On the contrary, the preparation can be studied any number of times without its undergoing any change. In macroscopic dissection one cannot always determine with absolute certainty whether an observed fibre-like formation is a nerve branch or something else, without subjecting it to a histologic control examination. In the method advocated here, a complete serial section of each nerve branch in its whole extent always is obtained. Moreover, if a nerve branch penetrates into firm formations of connective tissue, as is the case e.g. with the articular nerves, it must obviously be very difficult macroscopically to follow the relatively fragile nerve branch for any considerable distance; here again the microscopic anatomical procedure is decidedly superior.

For studying the nerves of the shoulder joint, serial sections were made of three human fetuses, 77.5 mm., 74.8 mm. and 73 mm. in length, as well as serial sections of the right shoulder region from a fetus 215 mm. in length. All the material was impregnated with silver nitrate before sectioning, according to a previously described technique (Wrete[35]). It is a well-known fact that the peripheral nervous system attains its full development at a very early stage; thus, at the 20 mm. stage the different nerve branches,

broadly speaking, have already reached their terminal positions. It might therefore be expected that in the fetuses used here the peripheral branches to the joints, etc., had long since been formed, an assumption that was borne out by the observations made.

The nerves in two of the joints (74.8 mm., right shoulder joint; 77.5 mm., right shoulder joint) are represented in Figs. 3, 4, 6 and 7 by profile reconstructions. The individual nerves, as also the contours of the articular capsule, have been projected to a plane passing, in the longitudinal direction of the trunk, through the middle of the glenoid cavity and the middle of the head of the humerus. The approximate position of the plane (P—P) is schematically indicated in Fig. 1.

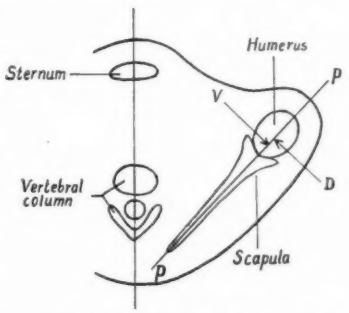

FIG. 1. Semi-schematic transverse section through the shoulder region of a human fetus. (P—P) Plane through the middle of the head of the humerus and through the scapula. (V) Supposed line of sight in profile reconstruction of the anterior side of the joint. (D) Supposed line of sight in reconstruction of the posterior side of the joint.

In Figs. 6 and 7 the joint is supposed to be viewed from the posterior side and at the same time somewhat from the lateral side (Fig. 1, D), in Figs. 3 and 4, from the anterior side and at the same time somewhat from the medial side (Fig. 1, V); thus, in all, four profile reconstructions have been executed. The other three joints were studied by finding and following the individual nerves in the serial sections, with the aid of the reconstructions.

The reconstructions, which were made with a magnification of 27.5, are not claimed to be perfectly exact in regard to the form of the joint or of the individual nerves, but only as regards the origin, course, ramification and distribution of the articular nerves, which in this case are the essential features.

In describing the distribution of the nerves in the joint, it seems natural, in view of the way in which I had proceeded in the study, to speak of the posterior side and anterior side of the joint, but it is, of course, also possible to combine the upper halves of these two sides and term it the "upper side" of the joint and the lower halves its "under side." One might also speak of an anterior upper quadrant, an anterior lower quadrant, and so on. That part of the articular capsule lying closer to the scapula, I designate as "scapular," and that lying closer to the humerus, as "humeral."

RESULTS

The articular nerves in each of the joints examined issue in a considerable number, varying between 9 and 12. They issue, in a way that varies somewhat in details, from four of the branches of the brachial plexus, namely the axillary, suprascapular, musculocutaneous and subscapular nerves, proceeding either from their main stems or from branches.

FIG. 2. Nerves to the anterior side of right shoulder joint according to Rüdinger. (1) Axillary nerve; (3) upper branch; (4) lower branch.

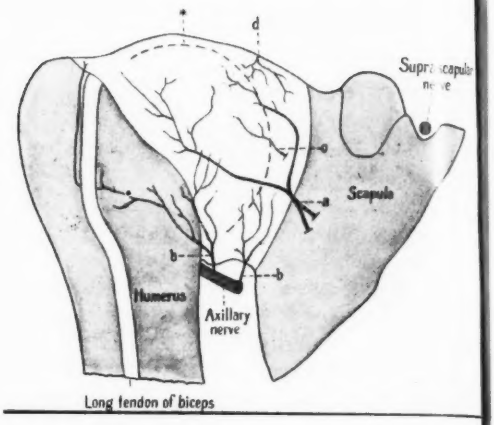

FIG. 3. Profile reconstruction of the anterior side of right shoulder joint in a human fetus 77.5 mm. in length. (*) Profile line of humerus head. (a) Articular nerve from the axillary and the musculocutaneous nerves. (b) Articular nerves from the axillary nerve. (c) Articular nerve from the subscapular nerve. (d) Branch of articular nerve from the suprascapular nerve.

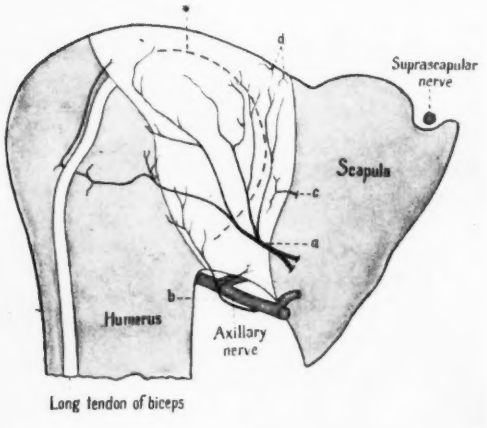

FIG. 4. Profile reconstruction of the anterior side of right shoulder joint in a human fetus 74.8 mm. in length. For notations, see Fig. 3.

FIG. 5. Nerves to the posterior side of right shoulder joint according to Rüdinger. (5) Suprascapular nerve; (6) upper branch; (8) lower branch.

Rüdinger's view that the anterior side of the joint is supplied by an upper and a lower articular nerve from the axillary nerve (Fig. 2) has, broadly speaking, been confirmed, but with the reservation that the upper nerve (Fig. 3, a and Fig. 4, a) receives an additional branch from the musculocutaneous nerve, and with the addition that the lower nerve may be replaced by two independently issuing nerves. It has been found that the anterior

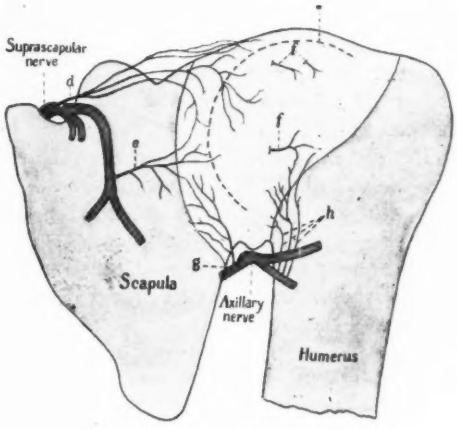

FIG. 6. Profile reconstruction of posterior side of right shoulder joint in a human fetus 77.5 mm. in length. (*) Profile line of humerus head. (d) Upper branch of the suprascapular nerve. (e) Lower branch of the suprascapular nerve. (f) Branches from nerves of supraspinatus and infraspinatus muscles. (g) Axillary branch to scapular part of capsule. (h) Axillary branches to humeral part of capsule.

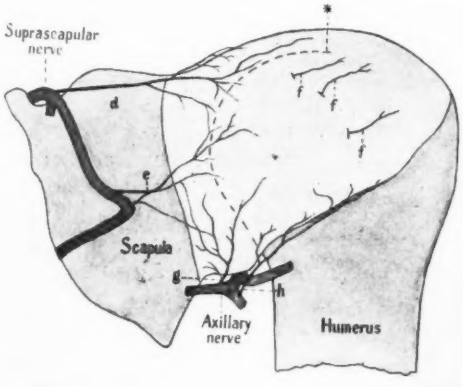

FIG. 7. Profile reconstruction of the posterior side of right shoulder joint in a human fetus 74.8 mm. in length. For notations, see Fig. 6.

side of the joint also receives branches from the subscapular nerve, running within the tendon of the subscapular muscle (Fig. 3, c and Fig. 4, c).

As for the posterior side of the joint, Rüdinger's description (Fig. 5) can be confirmed insofar as the suprascapular nerve, with an upper and a lower branch, takes part in its innervation (Fig. 6, d and e; Fig. 7, d and e).

It must be added, however, that in the shoulder joints examined in the present study, the axillary nerve, with a varying number of branches, also supplies a considerable part of the posterior side (Fig. 6, g and h, Fig. 7, g and h). The latter, finally, receives a number of branches from the suprascapular nerve, running within the supraspinatus and infraspinatus muscles and proceeding within those muscles from their own nerves (Fig. 6, f and Fig. 7, f).

As in many other parts of the peripheral nervous system, we see that an articular nerve may extend its terminal distribution and encroach on an adjacent nerve, which had been observed also by Rüdinger. Anastomoses may occur between adjacent articular nerves (in Fig. 6 between e and g; and between f and h; in Fig. 7 between e and g).

Besides the capsule and the ligament lying on its outer side, the articular nerves also supply the glenoidal labrum with numerous branches. I have not observed any branches to the articular cartilage, which, indeed, was not to be expected.

Rüdinger's description and illustrations give no clear information as to which of the articular nerves innervate that half of the articular capsule that lies nearest the scapula or the humerus, respectively. A study of the reconstructions shows that the scapular half of the capsule together with the glenoidal labrum is innervated on the anterior side (Figs. 3 and 4) at the top by the branch proceeding from the axillary and musculocutaneous nerves, below it by the subscapular nerve, and at the bottom by the axillary nerve. On the posterior side (Figs. 6 and 7) the scapular half is supplied at the top by the uppermost branch of the suprascapular nerve, in an intermediate part by the lower branch of the said nerve, and at the bottom by the axillary nerve.

The humeral half of the joint is supplied on the anterior side at the top by branches from the axillary and musculocutaneous nerves, and towards the bottom by the axillary nerve. (An exception in this regard must be made for the left shoulder joint of the 77.5 mm. fetus, where the last-mentioned branch is well developed, replacing the upper branch in the humeral half of the capsule.) The posterior side is supplied at the top by the nerve branches running in the tendons of the capsule tensors on that side and issuing from the suprascapular nerve, and below by one or more branches of the axillary nerve.

Some of the articular nerves follow slender blood vessels for longer or shorter distances. A minor part of the terminal rami, in their course, are likewise closely connected with the blood vessels.

In the main stems of the articular nerves and in the larger branches one often sees a distinct division into two kinds of nerve fibres, corresponding with observations previously reported.[35,36] Beside the intensely impregnated blackish-brown fibre bundles, which may be regarded as the sensory components in the nerve, we find brownish-yellow fibre bundles, which evidently are the sympathetic component.

A more detailed description of these joint nerves will follow in an anatomical periodical.

DISCUSSION OF THE RESULTS

In this investigation of the nerves of the shoulder joint the accuracy of Rüdinger's dissections has been verified, but by the different technique adopted, it has been possible to follow more closely the distribution of the articular nerves made known by Rüdinger's investigation. Moreover, new nerves to the joint have been detected. The demonstration that, with this technique, it is possible to get further than before in the study of the origin, course and distribution of these articular nerves is one of the principal results of this investigation. This warrants us in expecting that the same technique applied to other joints may enlarge our knowledge of the articular nerves there. Though it may be presumed that the articular nerves macroscopically detected by Rüdinger in the other joints are correctly described even if their distribution could not be followed in detail, we must also reckon with the possibility that, as in the shoulder joint, he may have overlooked certain nerves in those joints.

In view of the fact that the shoulder joint receives its nerves from four of the branches of the brachial plexus, namely the axillary, musculocutaneous, suprascapular and subscapular nerves, it seems possible, knowing what nerve segments take part in the constitution of those nerves, to form some idea as to what segments may conceivably participate in the sensory innervation of the shoulder joint. If we consider and compare the results of the analyses of the brachial plexus and the nerves branching from it that have been made by Herringham,[10] Wichmann,[29] v. Schumacher[25] and Kerr,[12] we arrive at the conclusion that the said nerves are made up of components from the following segments (enumerated in the order in which they are most frequently represented): the axillary nerve—C5, C6, C7, the musculocutaneous nerve—C5, C6, C7, C4, the suprascapular nerve—C5, C6, C4 and the subscapular nerve—C6, C5, C4, C7. In rare cases segments C8 and T1 also occur, except as regards the suprascapular nerve. These branches of the plexus thus together represent the segmental region C4–C7, and it is therefore theoretically possible that the sensory tracts for the shoulder joint pass through one, two or more of the dorsal roots of those segments. That any great importance can be attached to C4 in the formation of the brachial plexus is to some extent gainsaid by the fact that it takes part merely with a minor branch. From Bolk's[2,3] well-known anatomical investigations into the distribution of the sclerozones in the skeletal parts included on the shoulder joint, it is evident that the head of the humerus is composed mainly of parts of C6 and (to judge by the illustrations), to a minor degree, also of C5 and C7. The glenoid cavity of the scapula falls mainly within C6 but also, to a minor extent, within C7. The facts seem to indicate that the shoulder joint is formed mainly by C6, though also, to a minor degree, by C5 and C7.

In this connection it should be noted that Mårtensson,[16] by studies of

referred pain in the shoulder muscles in cases of arthrosis deformans in the shoulder joint and after experimental injection of hypertonic saline solution into the articular cavity in man, has indicated that the conduction of pain from the shoulder joint passes through one of the segments C5 or C6, or possibly both.

Thus, it is not possible to arrive at any definite view as to which segment or segments supply the shoulder joint: we must content ourselves with noting that C5, C6 and C7 are probably concerned. Nor is it possible to determine whether one, two or all three of these segments participate. It is also conceivable that C8 and T1 may be able to take part in exceptional cases.

There is also the question as to which parts of the sympathetic trunk may be conceived to send postganglionar sympathetic fibres to the shoulder joint. This will be briefly discussed, proceeding from the assumption that these tracts, as stated by White and Smithwick,[28] have a segmental course.

The plexus roots C4–C7 receive sympathetic fibres through the rami communicantes grisei coming from all the three cervical ganglia, i.e., the superior, the middle and the inferior cervical ganglion (Wrete[33]). As the latter ganglion is normally included in the stellate ganglion, being its cranial part, we must reckon with the possiblity that the shoulder joint, more precisely expressed, may receive its supply of sympathetic fibres from the region extending from the lower part of the superior cervical ganglion to the upper part of the stellate ganglion, or from a certain part of this sympathetic-trunk region.

The accessibility of the shoulder joint nerves for operative purposes now will be theoretically discussed. The articular nerves proceeding from the axillary nerve or from the origin of the axillary branches to the muscles issue at the medial axillary space, which seems to be, comparatively speaking, an easily accessible point. The same remark applies to the branches proceeding directly from the suprascapular nerve. It seems somewhat more difficult to find the branch proceeding from the axillary and musculocutaneous nerves, owing to its course higher up in the armpit, passing partly through its fat tissue. Lastly, I particularly desire to point out that some of the nerves to this joint are so situated that it seems to be extremely difficult, if not practically impossible, operatively to expose and excise them. I am referring to those nerves that proceed from within the capsule-tensing muscles on the anterior and posterior sides of the joint and pass to it through the tendons of those muscles. Therefore, even if all the other articular nerves were successfully excised, there would remain on the anterior side a region in the part of the capsule situated nearest the scapula, and on the posterior side in the part thereof situated nearest the humerus, in which some sensory tracts would still be intact.

SUMMARY

1. By studying in fetuses the Ag-impregnated articular nerves in histologic serial sections, their course and distribution can be followed more successfully than had previously been possible by means of dissection.

2. The shoulder joint in man receives a fairly large number of articular nerves (in the five joints examined numbering 9–12), coming partly from the main stems, partly from the branches of four nerves proceeding, in turn, from the brachial plexus, namely the axillary, suprascapular, musculocutaneous and subscapular nerves.

3. The anterior side of the joint is supplied from the axillary, musculocutaneous and subscapular nerves, its posterior side from the suprascapular and axillary nerves.

4. The scapular half of the joint (inclusive of the glenoidal labrum) is supplied from the axillary, musculocutaneous, suprascapular and subscapular nerves. Its humeral part is supplied from the same nerves, except the subscapular nerve.

5. An articular nerve may encroach on the area of distribution of an adjacent articular nerve.

6. Slender anastomoses between adjacent articular nerves may occur.

7. In view of the origin of the shoulder-joint nerves from certain branches of the brachial plexus, it may be inferred that the sensory tracts which pass through them from the joint go mainly through one, two or more of the plexal roots C4–C7 and the corresponding dorsal spinal nerve roots.

8. A possible starting point for the postganglionar sympathetic fibres, which in all probability exist in the articular nerves, is presumably a rather considerable part of the sympathetic trunk, notably a caudal part of the superior cervical ganglion, as well as the middle cervical ganglion and a cranial part of the stellate ganglion.

9. Only a small number of articular nerves are so situated that they cannot be operatively reached and excised without at the same time sacrificing motor tracts.

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RESTORATION OF CEREBRAL FUNCTION AFTER PROLONGED CARDIAC ARREST*

JAMES C. FOX, JR., M.D.

Department of Neurology-Neurosurgery, Hartford Hospital, Hartford, Connecticut

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SUDDEN stoppage of the heart during an operation has always been a terrifying experience for the surgeon and anaesthetist. A busy surgical clinic can anticipate an average of one or two cases of cardiac arrest a year. Adequate preparation and prompt determination to meet such emergencies when they arise may prevent otherwise certain fatalities. In recent years effective methods of treating ventricular fibrillation have been developed.[2,3,4,5] The procedure most recently recommended by Beck *et al.*[4] includes exposure of the heart, injection of procaine, cardiac massage and electrical stimulation.

If such individuals are to be saved in greater numbers in the future, what about the risk of cerebral damage from prolonged anoxia? The nerve cells of the brain are more sensitive to oxygen lack than are any other cells of the body. A generally accepted dictum has been that the human brain will not stand more than five minutes of oxygen deprivation without suffering irreparable damage. The following case, already reported,[8] is an example of reversible changes due to cerebral hypoxia during artificial maintenance of the circulation and respiration over a period of 27 minutes of ventricular fibrillation.

CASE REPORT

J.R., a 7-year-old boy, was admitted to the Hartford Hospital July 17, 1947 with a deep laceration of the sole of the right foot. There had been only moderate loss of blood. The child showed no sign of shock. Preoperative medication consisted of morphine sulphate gr.1/12, atropine sulphate gr.1/200 and seconal gr.3/4. The operation was performed under anaesthesia produced by open drop ether induced with cyclopropane. Cleansing and irrigation of the wound and repair of the tendons, muscles and nerves required about 75 minutes. At the close of the operation, 5 minutes after terminating the anaesthesia, the pulse suddenly stopped. The child's color became ashen, no heart sounds could be heard and respirations ceased. Approximately 2½ minutes from the time the heart stopped, the surgeon, Dr. R. S. Lampson, made an incision into the left 5th interspace, requiring 10 seconds to open the chest. The heart felt soft and lifeless but fine ripples of fibrillation were seen passing over the left ventricle beneath the pericardium.

Cardiac massage was immediately started and continued by intermittent compression of the heart against the sternum with the middle and index fingers of the left hand at a rate of 60 times per minute. A pulse could be felt at the wrist with each compression of the heart. The anaesthetist maintained artificial respiration

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with the anaesthetic machine and supplied pure oxygen to the lungs. Spontaneous respirations began 15 minutes after the onset of cardiac arrest, while the heart was still being massaged but had shown no sign of contraction. These were amplified by compression of the breathing bag. Two intravenous injections of 1 per cent procaine hydrochloride were given, the first of 3cc. 18 minutes after cardiac arrest, the second of 2cc. 26.5 minutes after cardiac arrest. Thirty seconds after this the heart stiffened beneath the massaging fingers and began to contract spontaneously. After 2 minutes of irregular contractions there developed a rapid, strong, regular pulse.

An electrocardiographic record had been started 20 minutes after cardiac arrest. This indicated ventricular fibrillation with 365 complexes per minute interrupted by a period showing no cardiac activity. Following a series of ten bizarre, aberrant, arrhythmic ventricular contractions, some of which seemed to be preceded by P waves, a gradual return of the normal mechanism took place. Observation of the heart was continued for ½ hour with the thorax open and 100,000 units of penicillin dissolved in 10cc. of procaine were flushed onto the pericardium. The chest wound was then closed and a plaster boot applied to the foot.

The chronological sequence of events during the operative period has been tabulated as follows:

5:50 P.M.	Anaesthesia started
7:22	Anaesthesia stopped
7:27	Cardiac arrest
7:29½	Chest opened
7:30	Cardiac massage begun
7:42	Spontaneous respirations returned
7:45	3cc. of 1% procaine I.V.
7:47	Electrocardiogram started
7:53½	2 cc. of 1% procaine I. V.
7:54	Spontaneous heart action resumed
7:56	Cardiac rhythm normal
8:07	Penicillin and procaine onto pericardium
8:35	Chest closed

When the patient left the operating room 2 hours after the heart had stopped, he was comatose, restless, vomiting, coughing and breathing spontaneously. He was placed in an oxygen tent under the care of a special nurse. The child could be aroused from coma for the first time 10 hours after resuscitation. Two hours later his first words were spoken. Neurological survey the following morning revealed all limbs flaccid and moving purposelessly, deep reflexes diminished, extensor left plantar response (right foot in cast), pupils equal with active light reflex. He remained semi-conscious during the balance of the day with alternating fixed stare and roving movements of the eyes. He became increasingly restless, moaned, responded to spoon feeding and lapsed into periods of sleep with repetitive chewing movements of the jaws and lips, and was incontinent.

During the second 24-hour period he became progressively wakeful and talkative. By the evening of July 18 he could give his name; the next morning he told his home address and ordered breakfast. During the course of the day he conversed with imaginary playmates, thought he was chewing bubble gum, and often replied irrelevantly when questioned. He remained very restless, mentally confused and irritable. Neurological survey showed full visual fields to confrontation tests, faulty

ocular fixation with apparent loss of visual acuity, clumsiness of the hands associated with bilateral astereognosis, return of flexor plantar response on the left.

During the third 24-hour period, he continued to talk irrationally at times and was subject to visual hallucinations—mother, dog, cat. While asleep, there occurred one brief generalized convulsion.

Neurological examination was performed in two stages on July 21, 85–90 hours postoperative, when the child was alert and in good rapport. His span of attention was shortened and he tired rapidly. He was oriented in all spheres, gave age, birthday, name of school, grade and could add and subtract single numbers. His speech was clearly enunciated with occasional dropping of syllables but no other evidence of expressive disorder. He was unable to write his name but formed a few letters. This was interpreted as due both to dysgraphia and clumsiness. He refused to read and protested vehemently when urged to do so. The special senses of smell and hearing were intact. Visual acuity was grossly impaired but could not be tested quantitatively. Color vision was intact. The eyes moved conjugately and smoothly without evidence of ocular weakness or nystagmus. However, he closed his right eye to avoid diplopia, especially on lateral gaze. The pupils were in mid position, equal, and reacted actively both directly and consensually to light and on accommodation. The other cranial nerves were normal. Ophthalmoscopic examination was negative. There was no muscular weakness or tremor but a generalized diminution of muscle tone. His hands were clumsy in the attempted performance of skilled acts. This was believed due to astereognosis. He was unable to distinguish coins of different size or identify common objects, particularly in the non-dominant left hand. Crude sensation for touch and pin prick was not impaired. The deep reflexes were uniformly diminished; abdominal and cremasteric reflexes were active; left plantar response was flexor.

During the following 10 days the child gradually regained the use of his hands with full return of discriminatory sensation. Incontinence of urine disappeared. He became overactive, talkative, irritable, fretful, sarcastic, and at times impertinent. When asked to write his name he formed a few letters and then abandoned the effort. Drawing of pictures was more successful. He was frequently remarking, "I'm a dope," and "I wish I were dead." He enjoyed the comics but when asked to read complained, "I can't see" or, "I forgot how."

By July 31 he was able to feed and dress himself but was still somewhat clumsy. He was interested in trying simple forms of occupational therapy but his hand had to be guided in lacing due both to clumsiness and impaired vision. Visual acuity on this date was R 20–70, L 20–70, both 20–50 (–1). He read Yaegger No. 4 type. The visual fields, ocular muscles and pupils were normal.

Progressive recovery took place during the last 10 days in the hospital. He began to read and write but continued to hold the page close to his eyes. On August 6 he wrote a note, "Doc, how many needles. They stink." His disposition became more cheerful and he was interested in all that went on about him. On August 7 visual acuity had increased to 20–40, R and L. He went home on August 11, the 26th postoperative day, an alert and lively but rather spoiled child.

He was re-examined on Oct. 17, 1947, 3 months after operation on return to school (3rd Grade). The mother reported that his overactivity had gradually diminished during the past 2 months. However he was still impulsive, restless, irritable, resentful of correction, and at times negativistic and difficult to manage. She interpreted this behavior as an exaggeration of former traits noted by his teachers as

well as his parents. Neurological examination was essentially negative. He could read Yaegger No. 1 type but still preferred to hold the page near his eyes. The optic nerve heads were normal. There was no remnant of dysphasia, dyslexia or dysgraphia. His talk was a bit rambling; his span of attention was shortened; and he was restless and fidgety. He responded promptly and intelligently to questions and performed simple arithmetical calculations rapidly and accurately. No memory defect could be detected. On October 6, vision was 20–15, R and L. An intelligence test performed at school on October 14 had reported an I.Q. of 108 according to Form A of the "Otis Quick Scoring Mental Ability Test."

On Dec. 20, 1947, 5 months after operation, his mother considered him the "same boy we took to the hospital" and the problems of behavior similar to those present before the injury, due chiefly to his intense enthusiasms and excessive energy. He attended school regularly and was promoted to the 4th Grade in good standing. A Stanford Achievement Test performed in May, 1948 gave a final score of 3.8 as compared with 2.6 for May, 1947, prior to the operation. When examined at his home on July 17, 1948, on the anniversary of operation, he was found to be normal in every respect. EEG July 22, 1948 revealed an essentially normal pattern although the amount of slow activity, 6–7 per second frequency, was somewhat more than might be expected for a child of this age.

SUMMARY OF NEUROLOGICAL CHANGES

The 10-hour period of unconsciousness followed by short-lived regression of behavior to an infantile level are evidence of substantial brain injury. On the other hand, the capacity for complete recovery including final preservation of a high degree of intelligence, gradual disappearance of behavioral abnormalities, full restoration of personality traits and normal EEG findings clearly indicate the reversibility of the brain changes. That the visual and sensory cortex were particularly vulnerable to the injurious anoxic effects is shown by the severe and relatively prolonged loss of visual acuity and the marked astereognosis. The prominence of visual agnosia and particularly alexia are also significant in this connection. The lesser degree of susceptibility of the motor cortex is indicated by the absence of muscular weakness and the transitory development of pyramidal reflex signs. There were no persistent signs of dysfunction of the cerebellum, basal ganglia, brain stem, spinal cord, cranial or peripheral nerves.

PATHOLOGICAL CONSIDERATIONS

Susceptibility to ischemia obviously differs in the various regions of the brain. The so-called "higher" centers are more vulnerable than the lower. Lennox, Gibbs and Gibbs[9] reported that their subjects became unconscious when oxygen saturation of blood in the jugular vein fell below 24 per cent whereas the lower centers continued to function. There is other evidence to support a decrease in the metabolic rate and also a gradient in vascularity as the neuraxis is descended from cerebral cortex to spinal cord.

Histological studies carried out by Weinberger *et al.*[13] upon the brains of cats subjected to circulatory arrest of 7.5 minutes showed that the motor and visual areas of the cortex sustained the earliest and most profound

damage. The olfactory, orbital and temporal regions of the cortex were the least susceptible. Laminae III and IV were more severely affected than I and II. Of the basal nuclei the order of vulnerability is as follows:—lateral geniculate, hypothalmus, thalamus, globus pallidus, caudate nucleus. In ischaemic experiments the basal ganglia are less affected than either the cortex or Purkinje cells of the cerebellum.

In recent years much more attention has been directed to reversible circulatory changes in the brain. Three vascular syndromes have been described by Scheinker[10]—"vasoporosis," "vasoparalysis" and "vasothrombosis." In reality they may be considered as phases of the same morbid process depending on its duration and severity. Either because of constricted or dilated vessels there is diminished movement of blood. Stagnation results in hypoxia both of the vessel wall and the parenchyma supplied by the vessel. Hypoxia alone may cause increased permeability of the capillary wall to both fluid and cells.

Sugar and Gerard[11] have shown that the effects of an abrupt interruption of cerebral blood flow are primarily due to anoxia, but hypoglycemia, hyperapnia and also increased extracellular potassium, induced by the anoxia, all contribute to the impairment of function. Other constituents of metabolism also play a role. The stasis in the circulation leads to local accumulations of CO_2 and other metabolites. Carbon dioxide is an active dilator. Further vascular dilatation results in increased permeability of the vessel wall, especially at the point of transition between capillary and venule. At this point oxygen tension is at its lowest and the endothelium most vulnerable to injury. Accumulation of fluid and blood cells in the dilated perivascular spaces retards the circulation even further, setting up a vicious cycle and increasing the hypoxia of the nerve tissue.

ANIMAL EXPERIMENTS

The effect of circulatory arrest on the various nervous centers has intrigued investigators for many years. The earliest recorded experiments on animals, those of Stenonis on fish and of Swammerdam on mammals in 1667, described the effects of occlusion of the abdominal aorta.[6] Ligation of the innominate and left subclavian, or the carotids and vertebrals was widely employed over the past century in the study of ischemia of the brain. These older methods gave inaccurate data because of the abundance of collateral circulation.

Two ingenious methods of studying the resistance of cerebral cells and neurones to lack of blood supply have recently been reported. Weinberger *et al.*,[12] by clamping the pulmonary artery of cats, arrested the entire somatic circulation. Kabat *et al.*[7] were able to obtain complete temporary arrest of the cerebral circulation without the use of anaesthetics. Two weeks after ligation of both vertebral arteries, the dog is given atropine to prevent vagal inhibition, the trachea is intubated orally, and a blood-pressure cuff about the neck is inflated to a pressure of 350 mg. Hg.

According to Weinberger *et al.* cardiac arrest lasting 3 minutes, 2? seconds causes permanent alteration in behavior and psychic function whereas after 6 minutes vision and sensation suffer some degree of permanent injury. On the contrary, Kabat *et al.* found apparent complete recovery of function after 6 minutes or less of cardiac arrest. Both groups of workers agree that 8 minutes or longer causes permanent severe damage to the brain.

After restoration of vital centers there ensues a period of hyperactivity. Coma may last for 24 hours after 4 minutes' arrest. Following coma a transition period lasts several days with gradual return of cerebral functions including righting mechanisms. Ataxia of cerebellar type is the most persisting neural dysfunction prior to recovery. Young animals are more resistant than adult but resistance is diminished to adult level at age of 4 months. Ability of the newborn to recover functionally is 400 per cent greater than the adult. The least trickle of blood can maintain activity of nerve tissue for a surprisingly long period.

THE TIME FACTOR

The total elapsed time of cardiac arrest (27 minutes) extended far beyond the accepted safety zone. It is obvious that the combination of measures to artificially maintain both the circulation and respiration and at the same time provide oxygen, saved the brain (and the heart) from irreparable damage. We have what amounts to a human experiment of circulatory arrest with brain ischemia for 2.5 minutes followed by a period of hypoxia lasting 24.5 minutes. Somewhat similar conditions prevailed in the cases of cardiac resuscitation reported by Adams and Hand[1] and Beck *et al.*[4] In both instances the duration of cardiac arrest, 20 and 35 minutes respectively, exceeded the limit of safety, established by animal experiments, but the patients survived their hypoxic experience without apparent brain damage.

It would appear, therefore, that clinical evidence is accumulating that the healthy human brain will stand the degree of hypoxia likely to be associated with the resuscitation of a heart undergoing ventricular fibrillation. In other words if cardiac arrest is *promptly* and *effectively* handled and the heart survives, it is unlikely that the brain of that individual will suffer irreparable damage. This is in striking contrast to cerebral anoxia of carbon monoxide poisoning and overdoses of anaesthetic and narcotic drugs. Here the pathological changes tend much more to be irreversible, characterized by widespread foci of necrosis and degeneration involving the corpus striatum.

SUMMARY AND CONCLUSIONS

1. A case is reported of cardiac arrest of 27 minutes due to proven ventricular fibrillation, effectively treated by cardiac massage, intravenous procaine, and adequate administration of oxygen.

2. The clinical course of recovery of cerebral functions is described in detail.

3. The prolonged loss of visual acuity and the marked astereognosis indicate that the visual and sensory cortex were particularly vulnerable to the injurious anoxic effects.

4. Final complete recovery is evidence of reversibility of the brain changes resulting from the prolonged hypoxia.

5. Both clinical and experimental data support the concept of a gradient of susceptibility as the neuraxis is descended from cerebral cortex to spinal cord.

6. The probable pathophysiology associated with interruption of cerebral blood flow is briefly discussed with reference to recent animal experiments.

7. It would appear that, if the cardiac arrest due to ventricular fibrillation is promptly and effectively handled, and the heart survives, it is unlikely that the brain of that individual will suffer irreparable damage from the degree of hypoxia occurring during the period of resuscitation.

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SYRINGO-ENCEPHALOMYELIA

DISCUSSION OF RELATED SYNDROMES AND PATHOLOGIC PROCESSES, WITH REPORT OF A CASE

HOMER D. KIRGIS, M.D., AND DEAN H. ECHOLS, M.D.

Section on Neurosurgery, Ochsner Clinic, and Department of Surgery, Tulane University School of Medicine, New Orleans, Louisiana

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THE formation of cystic cavities within the brain and spinal cord may be secondary to a multiplicity of pathologic processes, including thromboses and emboli of the arteries, inflammatory and neoplastic reactions, trauma, and various other disorders that may interfere with the normal metabolic activity of some portion of the central nervous system. The syndromes that usually have been termed syringomyelia, syringobulbia, or syringopontia seem to present a special problem in the formation of cysts within the central nervous system. These terms have acquired certain connotations relative to the origin, location and progression of the cystic lesions. They have customarily been used to designate a syndrome suggestive of a slowly expanding, centrally located, intramedullary lesion. The pathologic process, at least in the majority of cases that have been examined in detail, has consisted largely of the cyst, with evidence of gliosis in the adjacent tissue. However, certain investigators[7,17] have stressed the presence of areas of homogenization which they believe are sites of future cystic formation.

There is wide divergence of opinion regarding the mechanism of formation of the cyst and its fluid. Most investigators[2,3,5,14,16] have considered the cyst to be the product of relatively benign glial proliferation and necrosis of the involved tissue. The process has generally been postulated to originate by atypical multiplication and differentiation of cells, probably spongioblasts, in the vicinity of the deep portion of the posterior raphé of the spinal cord or the median raphé of the brain stem. Ependymal cells have been reported to form the lining of some syringomyelic cysts; others have apparently had no distinct epithelial wall. The presence of ependymal cells may, at least to some extent, be dependent upon the communication of the cyst with the central canal of the cord or with some remnant of the primitive neural canal. Regardless of the type of cells forming the wall of the cyst, the essential symptom-producing activity appears to be the gradual accumulation of fluid within the cyst with compression of neighboring tracts and nuclei. Although the original fluid of the cyst may be the result of necrotic tissue having undergone liquefaction, its volume probably is increased by transudation of fluid across the wall of the cyst or by the secretory action of the cells lining the cyst. One is impressed by the probable importance of the

latter process by the rapid re-accumulation of fluid in such cysts from which the fluid has been aspirated but in which no adequate, continued system of drainage has been instituted. The origin of syringomyelic cysts from what have been described as ruptured germinal epithelial cells has been suggested[1] but the pathophysiologic process involved is not clear.

It has been pointed out that anatomically the central portion of the spinal cord, the area in which the great majority of these cysts seem to originate, is particularly vulnerable to vascular disturbances.[3] This area is supplied by the terminal branches of the posterior and anterior spinal arteries and it has been postulated that generalized disease of the spinal arteries may result in greatest changes in this central area. Although some investigators[3,7] have reported hyalinization and other pathologic changes in the adjacent vessels, these changes have not been consistently in evidence in this type of cystic disease.

Hassin[7] concluded that syringomyelia and syringobulbia are manifestations of abiotrophy. At postmortem examination of the spinal cord of a patient with syringomyelia, he observed evidence of degeneration in the central area of the spinal cord at considerable distances from the cyst. These areas were described as zones of homogenization which represented a reaction of degeneration without evidence of previous glial proliferation.

A review of the literature reveals that processes, varying from cystic dilatation of the central canal of the spinal cord (which might be more correctly termed hydromyelia) to cystic degeneration within a glioma, have been discussed under the term *syringomyelia.* An insufficient number of cases that might be termed true syringomyelia and in which treatment consisted of establishing a permanent communication between the cavity of the cyst and the subarachnoid space has been followed to reach definite conclusions regarding the neoplastic significance of the proliferation of glial cells which has generally been reported to be present about the cyst. Puusepp,[11] however, described 2 such cases of patients who were living 4 years and 3½ years, respectively. Each was still free of evidence of recurrence of compression by the cyst of adjacent neural structures. The same may be said in regard to reaching a final decision relative to the condition representing a progressive degenerative disease. Numerous patients have been observed to survive many years following acute manifestations of syringomyelia, finally succumbing to some intercurrent infection, which fact does not support the latter theory.

Most reports of postmortem examination of these patients indicate that cavitation appears to have begun in the region immediately posterior to the central canal and to have extended asymmetrically into the lateral and posterior funiculi and superiorly and inferiorly close to the base of the posterior columns of gray matter. Reports of the development and progression of neurologic defects in these patients support these observations and show that the lower cervical and cervicothoracic regions of the spinal cord are the most common sites of origin of the cysts. Multiple cysts have been

described[8] as well as a single cyst, which extended almost the complete length of the spinal cord.[9]

The preference of the syringomyelic process for the lower cervical or cervicothoracic region results in the common presenting complaint of diminution of normal sensation in the hands with weakness of the muscles of the hands and forearms and inability to make discrete movements with the fingers. The symptom complex most typical of syringomyelia includes a bilateral area of hypothermia and hypalgesia with reduced acuity of these sensations on only one side or more pronounced on one side below this level. The sensations of touch and pressure may be unaltered, and the term *dissociation of sensations* has been used to refer to this selective interruption of sensory paths. The acuity of proprioception and of the sensation of vibration may be greatly reduced. The sensations of touch and pressure survive more persistently, not only because of the greater distance of the anterior spinothalamic tract from the usual site of origin of the syringomyelic cysts, but because of the comparatively long ascending homolateral branches of the primary afferent neurons which mediate these sensations. Characteristically, motor manifestations of syringomyelia include spasticity and weakness of the muscles below the upper level of defective sensation. This may be confined to or considerably greater on the side opposite the more pronounced sensory loss. This neurologic picture, of course, is that of a deep lesion which has involved the fibers mediating the sensations of pain and temperature as they cross the mid-line and which has expanded asymmetrically to encroach upon the lateral and posterior funiculi. Invasion of the anterior columns of gray matter may be evidenced mainly by flaccid weakness or paralysis of the intrinsic muscles of the hands or of the muscles of the forearms.

In addition to these sensory and motor manifestations of syringomyelia, further evidence of the disease may consist of so-called trophic disturbances in the periphery and mutilating lesions which might be expected in the absence of adequate pain and temperature sensibility.

If the lesion is confined to the lumbar region of the spinal cord, the motor deficiency may consist of a flaccid weakness of some of the muscles of the lower extremities with evidence of an upper motor neuron type of lesion affecting the muscles supplied by more inferior segments of the cord.

Syringobulbia and syringopontia may present a considerably more inconsistent picture. Usually there is evidence of damage to several of the nuclei of the brain stem. The most commonly affected include the vestibular nuclei, the nucleus of the spinal tract of the trigeminal nerve, and the visceral and somatic efferent nuclei of the medulla. The fiber tracts most often sufficiently involved at this level to form a significant part of the clinical picture include the medial lemniscus, the descending tract of the trigeminal nerve and the solitary tract. Unilateral damage to these tracts results in reduction in the normal acuity of exteroceptive and proprioceptive sensations on the contralateral side inferior to the head and similar reduction in sensation over the homolateral side of the head. Usually in syringobulbia

and syringopontia, there is reduction of these sensations bilaterally over the head, trunk and extremities. The common variation in the degree of this hypesthesia suggests the usual bilateral asymmetry of the lesion. Evidence of the pathologic process causing clinically recognizable dysfunction of the autonomic centers and the pyramidal and extrapyramidal systems is less frequently seen. There is, however, no complex of symptoms that can be said to be nearly so characteristic for syringobulbia and syringopontia as that which often is presented by syringomyelia. In general, the signs and symptoms of syringobulbia or syringopontia are those of any glioma of the brain stem except that they may signify greatest involvement of structures relatively close to the floor of the 4th ventricle, such as the vestibular nuclei, the nucleus ambiguus, and the nuclei of the 5th, 6th, 7th, and 12th cranial nerves and of the long ascending conduction pathways.

The therapeutic measures for cysts of the brain stem and spinal cord, which at this time are considered of value, are incision and drainage, and irradiation. The latter has been reported to give varying degrees of relief. Frazier and Rowe[6] emphasized the importance of remembering that irradiation affects only the gliosis, and consequently is of little value in the presence of cavitation. It is generally conceded that such patients demonstrate most improvement following exposure of the cyst with incision and drainage.[4,5,6,10,12,13,15] It has been advocated that a foreign body be inserted into the cyst in such a way that the permanency of the communication between the cavity of the cyst and the subarachnoid space could be assured. Various substances such as dura mater, silk sutures, muscle, gutta percha and silver clips have been used. If the process is fundamentally benign and symptoms develop only because the continued secretory activity of the lining cells or some similar mechanism causes the slowly expanding cyst to exert undue pressure on adjacent structures, equalization of the fluid dynamics within the cyst and the subarachnoid space should effect a cure. It seems improbable that release of the contents of such a cyst into the subarachnoid space would ever overtax the cerebrospinal fluid absorptive mechanism. Obviously, the process of fluid accumulation within the cystic cavity is the major factor in precipitation of a severe phase of the disease. It is not evident that the proliferation of glial cells results in the formation of a tumor which alone is a significant factor in production of the various signs and symptoms characteristic of the disease.

The following case demonstrates the probable superiority of surgical treatment over irradiation therapy. It also proves the futility of merely establishing a communication between the cavity of the cyst and the subarachnoid space without taking measures to prevent its closure by the formation of scar tissue. Furthermore, it seems to represent a type of case for which the terms, syringomyelia, syringobulbia, syringopontia or syringoencephalomyelia, might logically be reserved. This would seem consistent in view of the fact that the essential pathologic condition in this case has been adequately demonstrated to consist of formation of fluid within the

central nervous system with accumulation of the fluid in a cyst within the substance of the central nervous system. Involvement of the brain stem by the cyst without communication with the ventricular system seems to exclude the possibility that this case might be classed as an example of hydromyelia, that is, cystic dilatation of the central canal of the spinal cord. The accompanying neurologic disorder seemed dependent upon gradual expansion of the cyst as more and more fluid accumulated.

REPORT OF CASE

E. G. S., a white boy, aged 12 years, experienced a brief attack of dizziness in June 1946. Two months later a second attack occurred while diving and was immediately followed by persistent soreness of the muscles of the neck. During the next 3 days the vertigo, sometimes associated with vomiting, became severe. Attacks could be precipitated or intensified by turning the head or eyes to the left. Dysphagia and hoarseness soon developed. When admitted to the hospital on Sept. 5, 1946, he also was complaining of a dull, aching pain in the occipital region, neck and shoulder on the left.

Examination revealed severe dehydration, inability to stand or walk because of generalized weakness and dizziness, horizontal nystagmus which was increased by lateral deviation of the eyes, especially to the left, paresis of the muscles of the pharynx, tongue, and larynx, and diminished acuity of proprioception in all extremities. Motor dysfunction was greater on the left in the case of each motor nucleus affected. There was no diminution of exteroceptive sensations in the distribution of either cranial or spinal nerves. Violent vertigo could be produced by slight degrees of active or passive rotation of the head. There was no papilledema.

The CSF pressure was 180 mm. of CSF. Cautiously applied jugular compression caused a slight rise of the fluid level without subsequent fall. The fluid was clear and the protein content was 325 mg./ 100 cc.

1st Operation. A ventriculogram, made on Sept. 9, 1946, revealed slight dilatation of the 3rd and lateral ventricles without displacement. On the same day, the posterior fossa of the skull was opened through a midline incision. Inspection of the floor of the 4th ventricle revealed an enlarged and cystic brain stem. The most pronounced thinning of the posterior wall of the cyst was at the medullopontine junction in the region of the left vestibular nuclei. Slightly xanthochromic fluid was aspirated from the cyst at this point through a No. 26 gauge needle. A defect approximately 0.5 cm. in diameter was made in the posterior wall of the cyst in this area. By cautious exploration with a small flexible rubber catheter it was determined that the superior extent of the cyst was at the mesencephalic-pontine junction. The inferior pole of the cyst was not palpated although the catheter was directed downward to the mid-cervical level. The arch of the atlas and the laminas and posterior spinous processes of the 2nd and 3rd cervical vertebrae were removed. The incision in the dura was closed with interrupted sutures.

Course. Ability to swallow was regained within 48 hours, when tube feeding was discontinued. The vertigo disappeared and the nystagmus decreased. Phonation rapidly became normal and sensory function became almost intact. During the 2nd postoperative week roentgen-ray therapy directed at the cyst was begun. However, on the 20th postoperative day the patient complained of dizziness and within a week his condition had deteriorated to the preoperative level.

2nd Operation. On Oct. 4, 1946, the previously visualized portion of the brain and cord were reexamined and the remaining cervical segments of the spinal cord were exposed. It was obvious that the cyst had re-expanded. The scar tissue closing the aperture between the cyst and the 4th ventricle was easily identified. An incision was made in the cyst at the level of the 5th cervical segment. A catheter was passed upward in the cyst from this point well beyond the previous opening. It was passed downward slightly beyond the midthoracic level without encountering the lower end of the cyst. It was apparent that the cyst involved mainly the posterior funiculi of the spinal cord and extended from some point inferior to the midthoracic level, superiorly to the junction of the pons and mesencephalon (Fig. 1).

A drain, made by twisting several tantalum sutures together, was inserted into the cyst and brought out through another opening, which was made about 2 cm. inferior to the previously made opening. The ends of the drain were sutured to the deep surface of the dura and the dura was closed tightly.

Course. The patient's 2nd convalescence was characterized by slow but steady improvement. Evidence of defective circulation of the CSF with inadequate absorption continued for several days. CSF was removed from the subarachnoid space in quantities of 30 to 50 cc. each day for a week. At the end of 2 weeks, the scalp over the craniotomy defect was pulsating normally and the symptoms began to clear more rapidly. The dynamics of the CSF as estimated by bilateral jugular compression became normal. It was evident that the intracystic fluid pressure had become equalized with that of the subarachnoid space undoubtedly as a result of the maintenance of the free transmission of fluid between the two spaces along the tantalum drain.

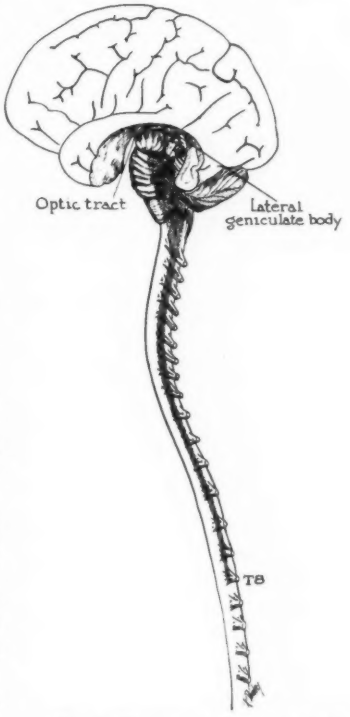

FIG. 1. Diagram of brain and spinal cord indicating extent and position of the encephalomyelic cyst in the case presented.

At the time of discharge from the hospital on the 24th postoperative day, the patient's greatest complaint was stiffness and weakness of the posterior cervical muscles. Evidence of neuropathologic changes consisted mainly of slight nystagmus with extreme abduction of the eyes.

The patient re-entered school shortly after returning to his home, and in spite of having missed almost 2 months, soon had completed sufficient work to enable him to continue with the regularly scheduled assignments. Re-examination after 6 months revealed no evidence of neurologic defects and the only complaint was a sensation of weakness of moderate degree after holding the head erect for relatively long periods. Two years following the 2nd operation, the patient was living a nor-

mally active life with no complaints and the neurologic examination yielded no evidence of pathologic changes. Roentgenography revealed the tantalum drain to have the same relation to the adjacent bony structures as immediately postoperatively.

DISCUSSION

In the case reported, syringo-encephalomyelia, consisting of a cyst extending from the mesencephalic-pontine junction to some point below the midthoracic portion of the spinal cord, was manifested clinically mainly by evidence of involvement of nuclear groups of the medulla as well as the tracts mediating proprioceptive sensation. Inspection revealed the cyst to occupy mainly the posterior funiculi of the spinal cord and the region immediately anterior to the 4th ventricle in the brain stem. Decompression by suboccipital craniotomy and upper cervical laminectomy plus excision of a portion of the wall of the cyst gave pronounced but temporary relief. Recurrence of symptoms followed closure of the defect in the wall of the cyst by scar tissue with re-accumulation of fluid and expansion of the cyst. Roentgen-ray therapy did not prevent rapid reappearance of the syndrome. Clinical recovery to date (2 years postoperatively) followed exposure of the cervical segments of the spinal cord and insertion of a tantalum drain between the cyst and the spinal subarachnoid space. With the drain in place and the dura intact, the cyst has the status of an extra ventricle in the system of intracranial and intraspinal fluid formation and absorption. With this relationship established, the cyst cannot exert undue pressure on adjacent nuclei and fiber tracts nor interfere with the normal circulation of cerebrospinal fluid. The prognosis continues to appear favorable after 2 years.

SUMMARY

The terms, syringo-encephalomyelia, syringomyelia, syringobulbia, and syringopontia have usually been used in a restricted sense to refer to what appears to be a pathologic entity consisting essentially of a slowly expanding cyst originating in a paracentral site in the spinal cord or brain stem. Most investigators have considered the cystic formation to be a manifestation of a neoplastic reaction, a developmental defect, or a degenerative process. Regardless of the exact etiology, the significant pathophysiologic activity of the disease is the accumulation of fluid within the cyst and the resultant compression of adjacent neural structures. The clinical manifestations depend upon the exact location of the lesion. The syndrome most characteristic of syringomyelia is indicative of a deeply placed lesion in the cervicothoracic region, involving first fiber tracts crossing or adjacent to the midline and later extending peripherally, usually in the posterior or lateral funiculi. Commonly, there is evidence of destruction of the nuclei of the anterior column of grey matter. Such a cystic lesion in the brain stem most often is manifested by neurologic evidence of damage to the nuclei and conduction pathways close to the floor of the 4th ventricle. Although irradiation therapy has been advocated, it has become more and more apparent that the

only satisfactory method of treatment is incision followed by some additional maneuver to insure permanent communication between the cavity of the cyst and the subarachnoid space. A case of syringo-encephalomyelia illustrating the superiority of surgical over irradiation therapy as well as the importance of establishing a permanent communication between the cystic cavity and the subarachnoid space is reported.

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FAILURE TO DISCLOSE RUPTURED INTERVERTEBRAL DISKS IN 32 OPERATIONS FOR SCIATICA*

DEAN H. ECHOLS, M.D., AND FREDERICK C. REHFELDT, M.D.†
Section on Neurosurgery, Ochsner Clinic, and Department of Surgery, Tulane University School of Medicine, New Orleans, Louisiana

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RECENTLY the orthopedic surgeons with whom we have been collaborating for the last 10 years on the problem of low back pain and sciatica made a follow-up study of 151 patients who had been treated surgically for these symptoms during a 5-year period.[1] They compared the results obtained by a neurosurgeon in cases of removal of ruptured lumbar intervertebral disks with the known results obtained in cases treated by disk removal combined with spinal fusion. The cases were rigidly selected by the orthopedists on the basis of conclusive demonstration at operation of a ruptured disk. Of the 151 cases, 34 were discarded as being equivocal herniations, non-disk lesions or negative explorations. The purpose of this paper is to present an analysis of these 34 cases.

TABLE 1

Classification of 151 cases of sciatica (1940–1944)

Classification	With Fusion	Without Fusion	Cases
Proved Disks	8	109	117
Other Causes	1	17	18
Negative Explorations	5	9	14
Cases Discarded	—	—	2
Total	14*	135	151

* Bone grafting was performed in approximately 9 per cent of cases during the 5-year period (1940–1944). During 1947 the percentage of combined operations had increased to 18 per cent. As a result of a follow-up study[2] of the 109 patients not having grafts the combined operation was discontinued by the orthopedists on January 1, 1948.

The 151 cases comprised all the private patients suspected of having a ruptured lumbar disk who had primary intraspinal explorations during the 5-year period ending December 31, 1944. The only patients excluded from this study were those who had had previous spinal operations elsewhere. All the operations were performed by one of us (D. H. E). In the combined operations the bone grafting was performed by an orthopedist.

* Read before the meeting of the American Academy of Neurological Surgery, Montreal, September 21, 1948.

† Fellow in Neurosurgery, Alton Ochsner Medical Foundation, New Orleans, Louisiana.

During our study of the 34 rejected cases (Table 1) it was found that 1 patient had had a previous spinal fusion elsewhere and should not have been included in the original group of 151 cases. In another case only the last name of the patient and the date of the operation are known. Consequently this study will be confined to the remaining 32 cases.

A study of the operative notes convinced us that in 18 of the 32 cases an adequate explanation for the sciatica was demonstrated at operation. These consisted of 8 cases of bulging disk, 4 cases in which the root was adherent to the disk, 4 cases of root compression by hypertrophy of bone, and 1 case each of spondylolisthesis and congenital malformation of the dural sac (Table 2).

TABLE 2

Equivocal herniations and non-disk lesions (18 cases)

Name	Age Years	Probable Etiology	Myelogram	Spinal Fusion	Rhizotomy	Second Operation	Follow-up Months	Results
1. Mr. E. B.	37	Bulging Disk	Positive	No	Yes	No	102	Excellent
2. Mr. R. H.	53	" "	None	No	No	No	68	Satisfactory
3. Mr. E. L.	27	" "	Positive	No	No	No	49	Excellent
4. Mrs. J. M.	42	" "	None	No	No	No	3	Unsatisfactory
5. Mr. A. M.	42	" "	None	No	No	Rhiz.	36	Satisfactory
6. Mr. L. M.	31	" "	None	No	No	No	43	Satisfactory
7. Mr. K. R.	48	" "	Positive	No	Yes	No	82	Satisfactory
8. Mr. D. S.	31	" "	None	No	No	No	83	Satisfactory
9. Mr. M. H.	36	Adherent Root	None	Yes	No	No	59	Satisfactory
10. Mr. O. R.	39	" "	None	No	No	No	49	Satisfactory
11. Mr. A. S.	56	" "	Positive	No	Yes	Rhiz. Fusion	52	Unsatisfactory
12. Miss E. W.	50	" "	None	No	Yes	No	76	Satisfactory
13. Mr. O. B.	36	Bony Hypertrophy	None	No	No	No	64	Satisfactory
14. Mr. O. B.	44	" "	None	No	No	Rhiz.	42	Unsatisfactory
15. Mr. E. M.	45	" "	None	No	No	No	4	Satisfactory
16. Mr. A. M.	65	" "	None	No	No	No	68	Satisfactory
17. Dr. E. S.	56	Spondylolisthesis	None	No	No	No	54	Satisfactory
18. Mrs. T. M.	35	Dural Deformity	Large Dural Sac	No	No	No	65	Unsatisfactory

The disk was opened in only 5 of the 8 cases of bulging disks. One of the 8 patients (Case 4) was not relieved by the operation and it is possible that another disk lesion was overlooked; she declined to return to us, became extremely depressed and committed suicide a few months after operation. The remaining 7 have had excellent or satisfactory results* for periods ranging from 35 to 90 months.

In all 4 cases of sciatica attributed to adhesions about the nerve root, it was necessary to dissect the root from the intervertebral disk, which was

* The functional results were tabulated as excellent, satisfactory or unsatisfactory. Results were considered *excellent* if the patient was in complete agreement with the examiner that there was absolutely no disability resulting from the original pain in his leg or back or from the operation performed. Results were classed as *satisfactory* if the patient, in spite of minor difficulty with his back or leg, returned to working full time without disability. *Unsatisfactory* results were obtained by those patients who could not work full time because of persistence or recurrence of pain in the back or leg.

narrow and hard but not bulging. We assume that this adhesive process is in some way the sequel to long-standing disk degeneration. Rhizotomy was done in 2 cases. Of these 4 patients 1 (Case 11) was not helped materially by rhizotomy and eventually had another rhizotomy and spinal fusion elsewhere without benefit. His result is classified as unsatisfactory, because even though he earns a good living as a tax expert, he does his work in bed and continues to complain of pain in the back and leg. The other 3 patients had satisfactory results after 49 to 77 months.

In 4 cases the root was found to be compressed by an apparent overgrowth of bone with resulting narrowing of the intervertebral foramen.[3,4] In each case the root was decompressed by facetectomy. Although hypertrophy of bone is a somewhat vague explanation for sciatica, the lesions were convincing and the follow-up study tends to verify the diagnosis. The period of follow-up study ranged from 4 to 69 months. Three of the patients had satisfactory results and 1 (Case 14) is considered unsatisfactory.

One patient (Case 17), a surgeon with spondylolisthesis and unilateral sciatica, requested an exploration in preference to spinal fusion. At operation the unattached 5th neural arch was removed. The 5th root was incarcerated in its foramen as the result of anterior dislocation of the 5th body. The root was decompressed by removal of the articular processes. The disk was not ruptured. This patient has had no further symptoms. Finally, exploration in a 41-year-old woman (Case 18) with a bizarre history, revealed nothing more than a dural sac which was twice the normal size, as had been demonstrated by myelography. In the last 2 cases (spondylolisthesis and malformation of the dural sac) the preoperative diagnosis was not necessarily herniated nucleus pulposus but exploration was carried out with this possibility in mind.

During this 5-year period no cases of cauda equina tumor simulating the disk syndrome were encountered. However, since this study, we have performed a negative exploration for a disk in a patient who eventually had an intradural neurofibroma removed elsewhere.

DISCUSSION OF EQUIVOCAL HERNIATIONS AND NON-DISK LESIONS (18 CASES)

It is possible that the apparent pathologic condition in a few of these 18 cases of miscellaneous lesions was not the actual cause of the sciatic pain. One or two of these patients may have had root compression well out in the intervertebral foramen, a cauda equina tumor, or a perineural cyst within the sacral canal.[5] However, the latter two possibilities seem somewhat unlikely in view of the fact that the follow-up period in the 17 living patients averages 57 months. It is, of course, impractical to discuss the results in a group of miscellaneous lesions but it can be seen that the finding of pathologic lesions other than disk nodules and free fragments of cartilage does not necessarily give a poor prognosis, for 14 of the 17 living patients are working full time even though they may not be free of pain in the back and leg.

NEGATIVE EXPLORATIONS FOR RUPTURED DISK (14 CASES)

The 14 cases in which no pathologic condition could be demonstrated at operation represent a problem concerning which there is universal controversy. Do most negative explorations represent inadequate exploration of the disk or unrecognized intradural lesions? Does a satisfactory negative exploration of the spinal canal in the lower lumbar region indicate root compression within the intervertebral foramen or within the sacral canal? Or is there a type of inflammatory or toxic radiculitis that can simulate the disk syndrome?

Although close scrutiny of our 14 cases of negative explorations (Table 3) has not provided the answer to these questions, it has stimulated our interest in the problem and has led to the formation of a more definitive program for the future.

TABLE 3

Negative explorations for ruptured disk (14 cases)

Name	Age Years	Probable Etiology	Unilateral Sciatica	Myelogram	Spinal Fusion	Rhizotomy	Second Operation	Follow-up Months	Results
19. Mrs. J. F.	45	?	Yes	Normal	Yes	No	No	78	Excellent
20. Mr. W. M.	40	?	Yes	Normal	Yes	No	No	69	Excellent
21. Mr. W. M.	37	?	Yes	None	Yes	No	No	65	Excellent
22. Mr. S. W.	38	Disk	No	None	Yes	No	No	66	Excellent
23. Mr. J. G.	30	?	Yes	None	Yes	No	No	16	Excellent
24. Mr. H. K.	36	Disk	Yes	Normal	No	Yes	No	38	Unsatisfactory
25. Mr. C. M.	30	Bony Hypertrophy	Yes	None	No	No	No	46	Satisfactory
26. Mr. R. A.	33	?	Yes	Normal	No	No	No	17	Unsatisfactory
27. Mr. W. H.	39	?	Yes	Normal	No	Yes	Rhiz.	0	Unsatisfactory
28. Mr. J. T.	40	Disk	Yes	Normal	No	Yes	Rhiz.	94	Excellent
29. Mr. T. C.	44	Disk	Yes	Normal	No	Yes	No	102	Satisfactory
30. Mrs. G. G.	35	Cord Disease	Yes	Normal	No	Yes	Rhiz.	51	Unsatisfactory
31. Mrs. C. G.	39	?	No	Normal	No	No	No	78	Unsatisfactory
32. Miss T. G.	46	Metastatic Carcinoma	Yes	Normal	No	No	Chord	75	Unsatisfactory

CASE REPORTS OF NEGATIVE EXPLORATIONS

Case 19. Mrs. J. F., aged 45 years, who had low back pain and unilateral leg pain which was not typical of root irritation, was rejected for operation on several occasions because of a normal myelogram. Eventually spinal fusion with negative exploration of the 4th and 5th disks was performed. She was promptly relieved of pain both in the back and leg and continues to be well 6.5 years later. We can offer no explanation for the leg pain in this case.

Case 20. Mr. W. M., aged 40 years, who had back pain and unilateral sciatica, was considered to be a candidate for a combined operation. We were confident that he had a ruptured 5th disk in spite of the fact that a myelogram made elsewhere was reported to be normal. The 4th and 5th disks were covered with normal veins. A probe was passed out through the corresponding foramens. The roots were neither swollen nor taut. Spinal fusion was performed. When last examined 69 months later, he was working full time as a laborer but complaining of occasional back pain. A settlement has been made between the patient and the insurance company. We are

at a loss to explain the sciatica in this case and it is possible that this patient had root compression which was not detected at operation.

Case 21. Mr. W. M., aged 37 years, complaining of back pain and vague pain in the hip and ankle, was subjected to exploration of the 4th and 5th disks without myelography at the time of a spinal fusion. No pathologic condition was disclosed. He was promptly relieved of symptoms and is still well 6 years later. We can offer no explanation for the pain in the extremity.

Case 22. Mr. S. W., aged 38 years, who had back pain and alternating sciatica, was considered to be a candidate for the combined operation. A myelogram was originally recommended but was not made for reasons no longer recalled. Exploration of the lower 2 disks gave negative results and spinal fusion was performed. A settlement was made with the insurance company. When last examined 66 months later, the patient was still completely free of leg pain and having only rare attacks of lumbago. A bulging of the disk near the midline may have been overlooked at operation.

Case 23. Mr. J. G., aged 30 years, who had back pain and bizarre pains in one thigh, was subjected to exploration of the disks without myelography at the time of spinal fusion. No pathologic condition was disclosed. He was completely relieved of symptoms but died of coronary occlusion 16 months later. We can offer no explanation for the pain in the extremity.

Case 24. Mr. H. K., aged 36 years, who had unilateral sciatica, was considered to have the typical signs and symptoms of a ruptured disk. A normal myelogram led us to conclude that a laterally placed rupture of the 5th disk was the probable diagnosis. The operation (April 1940) had to be discontinued because of uncontrollable bleeding. A week later another attempt was made to explore the 5th disk without success but the entire 5th root was severed. The patient still had some sciatic pain 3 months later when he returned to work. After 3 months of heavy labor he "reinjured" his back, became incapacitated, and made a settlement with the insurance company. Later he went to work in a shipyard in California and again became incapacitated. He was last seen by Dr. Carl Rand in Los Angeles in June 1943. At that time he was having a mild attack of sciatica. This is certainly a case of chronic recurrent sciatica due to rupture of the 5th disk. Unfortunately, the 5th root and not the 1st sacral root was sacrificed at the time of the two abortive attempts to explore the disks.

Case 25. (Compensation case) Mr. C. M., aged 30 years, had unilateral sciatica and back pain. In the process of extensive exploration of the lower 3 lumbar disks the 4th and 5th laminas were removed. The 5th root was unroofed well out into its foramen. No pathologic condition was demonstrated. The patient was relieved of symptoms and returned to his job. When examined 46 months later, he was beginning to have slight recurrence of sciatic pain. Although the radical exploration was negative, it is possible that we decompressed a root that was crowded by an unrecognized overgrowth of bone.

Case 26. Mr. R. A., aged 33 years, was injured by a fall in a shipyard and operated on 9 months later because of persistent low back pain and sciatica. Both the 4th and 5th disks were normal. The exploration was thorough to the extent that the veins overlying the disks were partially coagulated and removed and the intervertebral foramens were explored with a probe. The subsequent development of atrophy of the thigh and leg led to a myelogram which revealed no pathologic condi-

tion in the thoracic and lumbar regions. A settlement was made with the insurance company. Efforts to trace the patient have been unsuccessful but he is known to have been operating a taxicab 17 months after operation. We can offer no explanation for the back and leg pain in this case but are confident that it was not due to intraspinal rupture of a disk.

Case 27. Probably this case (Mr. W. H., aged 39 years) should not have been included in this study because the cauda equina was explored at the level of the conus in search of a tumor which was thought to be the cause of pain, weakness, and numbness in one lower extremity. No pathologic condition was demonstrated. The weakness and numbness disappeared after operation but the leg pain persisted. Six months later another neurosurgeon explored the lower lumbar disks and found "hypertrophied" ligaments. The 1st sacral root was severed. Since all efforts to trace the patient have failed, there is little likelihood of ever being certain of the etiology of the leg pain in this case.

Case 28. Mr. J. T., aged 40 years, presenting the typical signs and symptoms of ruptured disk, had a normal myelogram. An attempt to explore the 4th disk failed because of bleeding and the 5th root was severed extradurally. The patient was relieved of the sciatica and returned to heavy work. We reoperated on the patient 7 years later because of recurrence of the sciatica and back pain. This time the dura was opened and the sensory fibers of the 1st sacral root were severed. When last examined 7 months later, he was working, free of back pain, and complaining only of vague leg discomfort. We consider this to be a case of ruptured 4th disk which was not demonstrated at operation. However, there is some doubt as to this interpretation in view of the normal myelogram.

Case 29. (Compensation case) Mr. T. C., aged 44 years, complained of chronic low back pain and unilateral sciatica. A diagnosis of ruptured disk was made in spite of a normal myelogram. A normal, vein-covered 5th disk was disclosed at operation. Because of the absent left ankle jerk and the normal myelogram, the 4th disk was not explored. Instead, the dura was opened and the sensory bundles of the 1st sacral root were severed. The patient returned to his original occupation. When last examined 102 months later, he was beginning to notice slight pain in the other extremity. It is possible that this patient has a ruptured disk which was overlooked.

Case 30. Mrs. G. G., aged 35 years, a physician's wife, complained of pain in the back and thigh after a series of pararectal operations for severe pain in the hip. She had had 2 negative explorations with rhizotomies and repeated myelograms without finding any cause for her present painful state. A mild cord bladder suggests a degenerative lesion of the cord. The picture is complicated by a severe psychoneurosis.

Case 31. Mrs. C. G., aged 39 years, was thought to have a ruptured disk as the cause of bilateral sciatica but no pathologic condition was found at operation and the patient was not immediately improved. A subsequent myelogram was normal. Eventually she became completely free of back and leg pain for several years but when last seen 78 months after operation, she was suffering from the original symptoms. An explanation for her difficulties cannot be offered.

Case 32. Miss T.G., aged 46 years, in whom exploration was performed with reluctance and in spite of a normal myelogram, eventually proved to have metastatic carcinoma of the entire skeleton. The sciatic pain, which continued to be unilateral, was treated by chordotomy. She died 4 years after the original operation.

DISCUSSION OF NEGATIVE EXPLORATIONS (14 CASES)

We were eventually able to postulate a possible explanation for the sciatica in only 7 of our 14 negative explorations. Seven are still unexplained (Cases 19, 20, 21, 23, 26, 27, and 31).

Although the normal myelograms in 10 of the patients and the long follow-up studies in the remaining 4 patients practically rule out the possibility of tumor of the cauda equina in any of the cases, it is possible that in a few of our negative explorations there were disk protrusions or bony spurs far out in the intervertebral foramens.

Twelve of the 14 patients with negative explorations are believed to be alive. Of these all but 1 has been followed for periods ranging from 16 to 102 months. The average follow-up period is 60 months. We have conservatively classified the results in this group as excellent in 6 cases, satisfactory in 2 cases, and unsatisfactory in 6 cases.

COMMENT

This review of our experience with 14 negative explorations during a 5-year period has led to certain modifications of our policy as regards management of the disk problem. At present it is our belief that: (1) every candidate for operation on a lumbar disk should have complete orthopedic investigation before operation is discussed with the patient, (2) myelography should be performed in every case in which there is doubt as to the diagnosis of ruptured disk, (3) spinal fusion should not be done at the time of the disk removal, (4) exploration of the intervertebral foramens (facetectomy) should be carried out when no pathologic condition is found within the spinal canal, and (5) section of one or more sensory roots should be seriously considered in every case of negative exploration for ruptured disk.

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A SPECIFIC EPILEPTIC SYNDROME FAVORABLY AFFECTED BY LYSIS OF ANOMALOUS PACCHIONIAN GRANULATIONS

JOHN E. SCARFF, M.D.

Department of Neurosurgery, College of Physicians and Surgeons, Columbia University; and the Service of Neurological Surgery, Neurological Institute, Columbia-Presbyterian Medical Center, New York City

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In 1936 I described a specific epileptic syndrome favorably affected by lysis of anomalous Pacchionian granulations and made a preliminary report of 3 cases.[3] The present paper consists of a follow up of these original cases, with a report on 17 additional ones.

The essential features of this specific epileptic syndrome are:

1. *Early onset.* Convulsions usually begin during the first or second decade, suggesting a congenital lesion.

2. *Chronicity.* The cases here reported average more than 11 years in duration.

3. *Focal convulsions.* Characteristically these begin in either the shoulder or the lower extremity, the cortical centers for which are located near the superior border of the cerebral hemisphere. As the illness progresses, the focal nature of the attacks may be lost and the convulsions become generalized.

4. *Neurological examination negative.* This is true even in cases of long standing, thus excluding neoplastic or degenerative etiology. Slight pyramidal tract signs may be found in the affected limbs, but this is an inconstant finding.

5. *Pneumoencephalogram negative.* There is no characteristic or significant finding on air studies.

6. *Anomalous Pacchionian granulations.* This is the one abnormal finding on cerebral exploration. The granulations are larger than normal and impinge on the excitable motor cortex. The superior communicating veins which pass through them are larger and more numerous than normal.

7. *Lowered threshold to electrical stimulation.* This is a characteristic finding in the vicinity of the anomalous granulations. Whereas minimal stimuli yield focal responses at most points along the primary motor strip, the same stimuli in the vicinity of the anomalous granulations result in convulsions similar to the spontaneous ones.

The essential clinical feature of this syndrome is the existence of a congenital epilepsy of focal nature without objective neurological deficits. The presence of focal features sets this syndrome apart from the common, idiopathic epilepsy of infancy and childhood. The absence of demonstrable neurological deficits distinguishes it from other focal congenital epilepsies, such as those caused by birth injuries or gross developmental anomalies.

Twenty patients with syndromes approximating that just described were treated by lysis of the anomalous Pacchionian granulations and division of the superior cerebral veins passing through them.

Prior to operation, all these patients had received prolonged systematic anticonvulsant therapy without material benefit. Following operation, all of them were again placed on the same anticonvulsant medication they had received prior to operation. In no instance was the postoperative dosage greater than the preoperative one. After discharge, the patients were followed for varying lengths of time ranging from 5 months to 6 years. Com-

TABLE 1

Case	Age at Onset			Duration	Follow-up Time	Reduction in Frequency of Attacks
	1–10 yrs.	10–20 yrs.	20–30 yrs.			
1. C.G.*	1 yr.			18 yrs.	72 mos.	90%
2. S.G.*			23 yrs.	5 yrs.	48 mos.	90%
3. J.B.		14 yrs.		8 yrs.	30 mos.	90%
4. J.G.	3 mos.			4 yrs.	60 mos.	90%
5. A.G.	1 yr.			13 yrs.	28 mos.	90%
6. A.M.B.	5 yrs.			9 yrs.	28 mos.	90%
7. M.G.	6 yrs.			3 yrs.	15 mos.	90%
8. S.F.		15 yrs.		8 yrs.	30 mos.	70%
9. F.K.		17 yrs.		2 yrs.	18 mos.	70%
10. D.J.	18 mos.			4½ yrs.	30 mos.	70%
11. St.C.		13½ yrs.		1¼ yrs.	30 mos.	70%
12. S.B.	8 yrs.			4 yrs.	15 mos.	70%
13. E.M.		14 yrs.		3 yrs.	18 mos.	50%
14. B.L.		13 yrs.		22 yrs.	12 mos.	50%
15. C.W.		17 yrs.		2 yrs.	24 mos.	50%
16. F.A.	10 yrs.			5 yrs.	42 mos.	25%
17. M.S.*	2 yrs.			55 yrs.	5 mos.	25%
18. R.L.	2 mos.			11 yrs.	6 mos.	25%
19. W.S.			30 yrs.	1 yr.	24 mos.	0
20. A.K.		15 yrs.		35 yrs.	6 mos.	0

* Cases reported in preliminary communication, 1936.[3]

plete cure by this method is not claimed, but of the 20 patients here reported, 18 had the course of their illness favorably affected by the operation.

Table 1 shows the approximate reduction in convulsions in each of the patients. It will be noted that in 7 of the patients, the reduction in the number of attacks amounted to more than 90 per cent; in 5, it amounted to more than 70 per cent; in 3, it amounted to more than 50 per cent; in 3, it was more than 25 per cent; and in 2, there was no reduction in the number of seizures postoperatively.

CASE REPORTS

Case 1, C.G. This was a 19-year-old boy who had suffered from major convulsions for 18 years. In the beginning, these were limited to his left leg, but they soon lost all focal features. For many years he had averaged 1 grand mal seizure each

day. For the year immediately preceding operation, he had suffered 10 grand mal seizures daily, and when admitted to the hospital he had been in status epilepticus for 48 hours.

After operation, this patient was followed for 6 years, during which time he had not a single convulsion.

Case 2, S.G. This 28-year-old woman had suffered focal and generalized convulsions for 5 years before admission. The focal seizures involved only the right leg. During the year before admission, she had experienced 1 or 2 grand mal seizures nightly. For 7 weeks prior to operation, while receiving large doses of luminal on the neurological ward, she had suffered 2 generalized seizures each night.

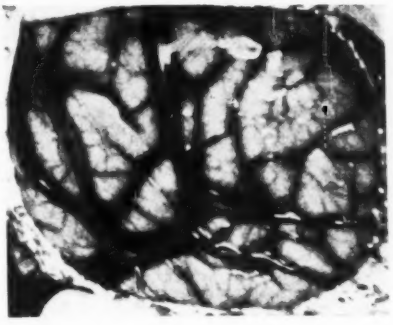

FIG. 1. Case 4. The large vein crossing the cortex leads to a dense, anomalous Pacchionian granulation on the superior border and superior mesial surface of the hemisphere. Surgical division of vein and granulation resulted in complete cessation of grand mal seizures during the 5-year period of postoperative observation.

After operation, she was followed for 4 years, during which time she had only 2 or 3 major attacks per year, all of which occurred during her sleep.

Case 4, J.G. This 4½-year-old boy began to have right-sided convulsions at the age of 3 months. These eventually became generalized and increased in frequency. For 15 months, while receiving phenobarbital and Dilantin to the point of toxicity, he continued to have 1 grand mal seizure each day. During the 10 days immediately preceding operation, he suffered 8 to 10 attacks each day. When admitted to the Neurological Institute, he was in a state of exhaustion.

During the 5 years since this boy was operated upon, he has not had a single convulsion, and during the past 4 years he has taken no medication (Fig. 1).

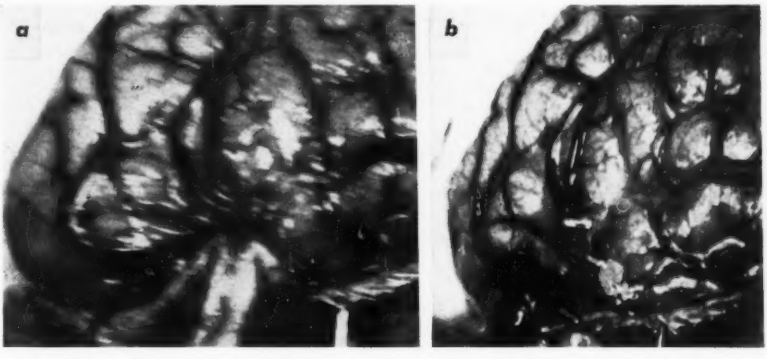

FIG. 2. Case 5. Anomalous Pacchionian granulation and venous plexus (a) overlying excitable motor cortex. Division of the veins and Pacchionian granulation (b) resulted in a 90 per cent reduction in attacks.

Case 6, A.M.B. This 14-year-old girl began to have left-sided convulsions at the age of 5. At the age of 7, these became generalized. From the time she was 8 until she was 14, she had averaged 4 to 5 grand mal convulsions per day. During the first week of her hospitalization here, there were recorded more than 10 major seizures each day. Twenty-four hours before operation, status epilepticus developed, uncontrolled by drugs.

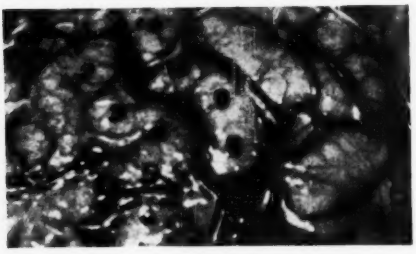

FIG. 3. Case 6. The brain here pictured is that of a child who suffered 4 to 5 grand mal convulsions daily for 6 years before operation. Following division of the large anomalous vein and its associated Pacchionian granulation, this child experienced only 3 attacks during the 2 years and 4 months of postoperative observation.

After operation, this patient was followed for 2 years and 4 months, during which time she experienced only 3 convulsions (Fig. 3.)

Case 10, D.J. This 6-year-old girl had had convulsions since the age of 18 months. At first, these were limited to the left side of the body. The attacks increased in frequency, and by the time she was 6, they were occurring about once each month.

In the 5 years that this patient was followed after operation, she had no attacks.

Comment. The extent of relief was not so great in all of the cases as in these 5, although improvement in varying degrees was attained in 18 of the 20 cases. Table 1 shows, together with other data, the degree of improvement in each case. It will be noted that 2 of the patients conforming to this syndrome were not helped by the operation. No explanation for this variation can be formulated.

A control group of 3 patients with generalized convulsions without the features of the specific syndrome herein described were operated upon as controls. All Pacchionian granulations for a considerable distance along the superior border, anterior and posterior to the central sulcus, were divided on both hemispheres. In none of these 3 patients was there any alteration in the incidence or severity of convulsions after operation.

DISCUSSION

In 1933, Ney[1] expressed the opinion that the traction of Pacchionian granulations upon the brain might be the cause of *epilepsy*. But "realizing the impossibility of separating these extensive cortical adhesions surgically," he concluded that "the removal of the vertex of the skull and the substitution therefor of a flat celluloid plate might serve to prevent stretching at these points by lowering the cortex."

In 1936, my paper[3] describing the *lysis* of Pacchionian granulations for relief of a *specific* epileptic syndrome was published. In 1939, Ney[2] wrote that his "present operative procedure has become more radical," and then included "separating cortico-dural attachments at the vertex when they can be demonstrated roentgenologically."

Whether or not the Pacchionian granulations have a physiologic role in the absorption of cerebrospinal fluid from the subarachnoid space has not yet been settled. It is certain, however, that they have a special mechanical function to which little attention has been paid: Pacchionian granulations hold the superior border of the hemisphere tight against the roof of the calvaria to protect the large, thin-walled superior cerebral veins in their passage from the surface of the brain to the superior longitudinal sinus. Each time a step is taken or the position of the head is changed, traction is exerted on the brain through these firm, unyielding bands of tissue. It is remarkable that this constant tugging on the brain does not produce convulsions more often, and this must indicate that in the normal brain an unusually high threshold-barrier to traction stimuli exists in the vicinity of these granulations.

In certain patients, however, the Pacchionian granulations may be anomalous in size or structure, or occupy an anomalous position on the surface of the brain in respect to the excitable cortex. In some cases, too, the brain itself may have a constitutionally low threshold for mechanical stimuli. Under such circumstances, the continuous traction of Pacchionian granulations may suffice to induce convulsions. In these cases, division of the anomalous granulations, with release of the brain from the rigid calvaria, should result in lessening of such attacks. The cases herein reported support this concept.

The thesis here offered does not demand that the anomalous Pacchionian granulations necessarily represent the primary etiologic factor in the production of the convulsions. It may well be that the granulations merely represent a secondary "mechanism of release" for convulsive phenomena in a constitutionally epileptic brain.

The alteration in venous drainage resulting from the incidental ligation of the veins passing through the granulations may be a factor in the postoperative reduction of convulsions. Transient hemiparesis or aphasia, due presumably to venous congestion, occurred in approximately $\frac{1}{3}$ of the cases, but in every instance had completely disappeared within 10 days after operation. Whether this alteration in circulation is an important factor in the reduction of convulsions is not clear.

SUMMARY

(1) A specific epileptic syndrome is described, which is relieved by lysis of anomalous Pacchionian granulations.

(2) Twenty cases conforming to this syndrome and treated in this manner are reported.

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CRANIOSTENOSIS

WITH NOTES ON A MODIFIED OPERATION FOR THE BRACHYCEPHALIC FORM

DAVID FAIRMAN, M.D.,* AND GILBERT HORRAX, M.D.
Department of Neurosurgery, the Lahey Clinic, and the New England Deaconess Hospital, Boston, Massachusetts

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CRANIOSTENOSIS is the premature synostosis of two or more membranous bones of the cranial vault. Although normally the sutures are apparently closed at the end of the first year because the serrated margins become interlocked, the x-ray examination reveals that the bones are not actually fused. During this period, growth of the skull is feasible because of the existence of the suture spaces. Later the skull growth is only by periosteal apposition and resorption, just as in basal synchondrosis. Ordinarily the brain doubles its weight in the first 7 months of life and triples its weight in 2½ years, completing 80 per cent of its entire growth in the first 3 years of life. For this reason craniostenosis has practical importance when it occurs during the intra-uterine period or first year of life.

Based on anatomical and clinical facts we have suggested the following classification:[4]

- Craniostenosis
  - Incomplete
    - Scaphocephaly
      - Compensated
      - Decompensated
    - Brachycephaly
      - Compensated
      - Decompensated
  - Complete — Oxycephaly
    - Compensated
    - Decompensated

Premature closure of the sagittal suture results in a long narrow skull (scaphocephaly) and early synostosis of the coronal suture develops a broad, short and high skull (brachycephaly). When premature synostosis affects practically all the sutures the result is a high peaked skull (oxycephaly).

Other changes in the skull may also appear. The anterior fossa becomes short, with its floor oblique, and the orbit becomes shallow, with prominence of the eyes.

Craniostenosis is compensated when a compensatory growth of the skull occurs at the open sutures. Since this is the only evidence of this condition, a markedly abnormal shape of the head results which must be corroborated by the roentgenologic findings. Craniostenosis is decompensated when the

* Formerly Fellow in Neurosurgery, Lahey Clinic, Boston, Massachusetts. Now at Sgo. del Estero 286, Buenos Aires, Argentina.

patient develops chronic increased intracranial pressure, as evidenced by headache, vomiting, mental deterioration and convulsive seizures. Papilledema and optic atrophy with blindness may occur. In these cases an air injection is advisable to rule out a tumor or some other intracranial lesion.

OPERATIVE PROCEDURES

The first attempts at surgical treatment of craniostenosis were made by Krause in Berlin and Eiselsberg in Vienna, performing subtemporal decompression in some cases of decompensated craniostenosis as a palliative measure for relieving the increased intracranial pressure.

The first surgical treatment with curative purposes was done by Lane[8] on August 28, 1888. He removed a strip of bone an inch wide, extending from the anterior to the posterior fontanelle. Then he removed on each side sections of the remaining parietal bones, and as Lane described it, the space after resection resembled a cross, of which the arms were of equal length and breadth. The child (9 months old) lived 14 hours after operation. Death was attributed to the prostrating effect of the triple alcohol-chloroform-ether anesthesia used. A year later he performed the same operation on another infant in whom the space after resection resembled a letter H; the results were better—the child survived and showed some mental improvement. Unfortunately, the 2 patients operated on by Lane were imbecilic infants with microcephaly, and consequently the procedure fell into disrepute.

More than 35 years elapsed until Faber and Towne[2] adopted Lane's operation. By this time, as we will see later, it was used as a preventive measure in oxycephaly and allied conditions.

Subtemporal decompressions for oxycephaly were performed in Cushing's clinic at least as early as 1913. One of these patients whose end result is known was operated upon by one of us (G. H.) on July 3, 1916. This was a 9-year-old girl who had decompensated oxycephaly, with bilateral exophthalmos, papilledema and headaches. She has been relieved of her pressure symptoms for more than 20 years.

Operations for craniostenosis prior to 1924 were performed only as palliative measures after increased intracranial pressure had developed and the usual irreparable damage had occurred. Although Messmer, in 1921, advocated extirpation of synostosed sutures, the operation was never performed. It was not until 1924 that Faber and Towne[2] advocated the revival of the Lane-Lannelongue operation to be performed sufficiently early in life to prevent the effects of premature synostosis. In June 1924, they performed a linear craniectomy in a child aged 9 months, who showed a rapidly progressing deformity of the skull due to synostosis of the sagittal and right coronal sutures, with signs of increasing intracranial pressure. Two channels of bone, 1 cm. wide, were removed by means of a transverse scalp incision. The first was longitudinal, extending from the coronal to the lambdoidal suture and to the right of the sagittal suture, and the second extending from one squamoparietal suture to the other, posterior to the coronal sutures.

The result of the operation was excellent, the shape and development of the head becoming approximately normal. Considering the fact that growth of the skull and brain is very rapid during early infancy, Faber and Towne for the first time advocated the operation during the first 6 months, before malformation has progressed too far and signs of decompensation have appeared.

In 1932, Bauer described an operation for oxycephaly consisting of a circular resection of the skull, leaving the upper segment of the skull free to rise beneath the scalp flap, thus relieving the increased intracranial pressure. Keegan,[1] in 1934, performed a bilateral subtemporal decompression in 2 stages, removing the bony channels as suggested by Faber and Towne, and opening the dura only on the right side. In 1935, Keegan performed a modification of Bauer's operation on another patient. Instead of doing a complete circular resection of the skull he spared a distance of about 3 cm. in the occipital region to permit some fixation of the skull cap.

King,[6,7] in 1936, devised an operation for decompensated oxycephaly, making a mosaic of the bones of the cranial vault. Fragmentation of the skull on each side created about nine fragments of bone by cutting between burr holes.

In 1943, Faber and Towne[3] reported 5 cases of compensated craniostenosis in which an early operation was done for the prevention of symptoms of increased intracranial pressure and sequelae which presumably would have developed. They modified their former surgical procedure, removing strips of bone about 2 cm. wide parallel to the closed suture. In the case of scaphocephaly the bony channels were removed on either side of the sagittal suture, connecting both defects through a strip of bone removed at a point just anterior to the lambdoidal suture. For brachycephaly they excised a strip of bone posterior to each of the closed coronal sutures, extending from the anterior fontanelle to the posterior squamous portion of the temporal bones. In oxycephaly they performed a transverse and longitudinal excision parallel to the closed coronal and sagittal sutures. Recently, Ingraham,[5] based on experimental observations, advocated the use of polyethylene to delay closure of artificial cranial sutures created by operation.

We were unable to find any mention in the literature of surgical procedures performed in cases of decompensated brachycephaly. The patients with brachycephaly operated on to date had compensated craniostenosis. An early operation was done to prevent the symptoms of increased intracranial pressure. Therefore, we consider it worth while to present the case report of a child who had a decompensated brachycephaly.

A 3-year-old white boy was admitted for the first time to the New England Deaconess Hospital on Nov. 14, 1947. He had had convulsions that appeared shortly after he was born. The convulsions were of generalized type with loss of consciousness lasting for about 2 minutes. He was able to walk at the end of his 1st year. He was able to speak a few words at 9 months but progressed no further. About 3 weeks before admission he lost the use of his legs. For 3 days he was unable to stand a

all gradually he regained the use of his left leg and in a few days his right one. At the time of admission he was able to stand and walk, but only a few steps. He had always had a peculiarly shaped head.

Examination. The neurologic findings were negative except for bilateral choked disks. Examination of the head revealed flattening in the anterior and posterior plane with the cranial wall abnormally high. The forehead was excessively prominent and the eyes were placed far apart because the root of the nose was widened. Macewen's sign was negative.

X-rays of the skull showed that the anterior fossa was shallow with marked convolutional impressions. The coronal suture was obliterated. The middle and posterior fossae were unusually deep, with the remainder of the skull appearing normal for his age (Fig. 1).

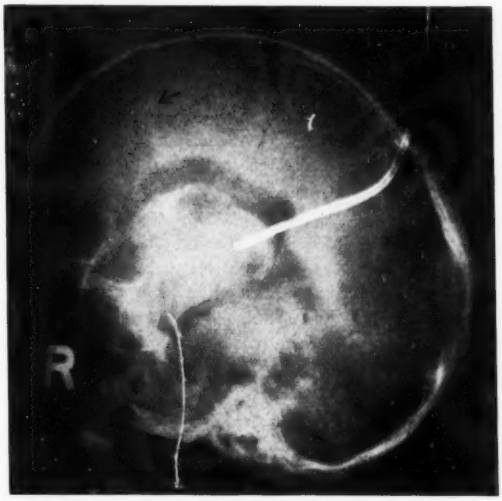

FIG. 1. Roentgenogram following air injection, showing obliterated coronal suture (arrow), normal lambdoidal suture and marked convolutional impressions in frontal region. The middle and posterior fossae are unusually deep.

On Nov. 20, 1947, ventriculography was performed. Approximately 10 cc. of clear CSF were obtained from each side and replaced by filtered air. The subsequent roentgenograms disclosed good filling of the ventricular system, which was normal except for some symmetrical dilatation of the frontal horns (Fig. 1).

From the clinical picture and roentgenologic findings the diagnosis of decompensated brachycephaly was made.

1st Operation. Following ventriculography a right frontotemporal parietal craniotomy was carried out. The dura was extremely adherent along the closed coronal suture, which was represented by a distinct white line through the bone, but there was no particular difficulty about turning up the flap. The bone was thickened in this suture area, not only above but particularly down toward the region of the

pterion where the bone made a much deeper projection inward than usual between the posterior end of the frontal lobe and the temporal lobe. A block of bone here was taken out where it indented the dura to such a great extent (Fig. 2 b), and also a considerable amount of bone was taken out in the temporal region from the lower portion of the bone flap and from the bone below the flap as well. Likewise, the area of the coronal suture superiorly was removed (Fig. 2 a), and the strip of bone which remained anterior, after taking out this piece, was replaced over the dura, leaving a gap in the bone between this strip and the bone anteriorly as well as between it and the anterior portion of the bone flap, so that expansion could take place here. Furthermore, the dura was opened over the temporal region where bone had been removed in order that a liberal decompression could be secured. The posterior and upper portion of the bone flap was then replaced, after which the skin and muscle flaps were replaced and careful closure carried out in two layers with silk, without drainage.

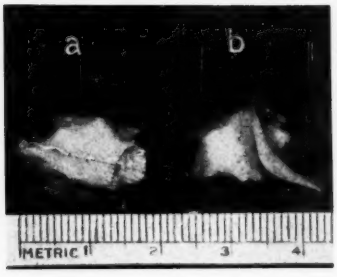

FIG. 2. (a) Segment of removed bone including portion of coronal suture. Note obliterated suture represented by white line. (b) Bone removed from region of pterion to show its abnormal depth.

The *postoperative course* was normal and the wound healed satisfactorily. By the time the patient left the hospital, there was marked improvement in his mental status as evidenced by the fact that he was less irritable, more alert and was taking nourishment better. Also, he was able to stand and walk merely by holding on to the edges of chair and table.

He was readmitted Feb. 20, 1948. At that time the child was alert and more cooperative. The gait was fair, but greatly improved as compared with that on his 1st admission. The decompression area was bulging and moderately tense. The optic disk margins still showed slight blurring, but there was no elevation.

Discussion. The head of this child was flattened in the anteroposterior plane and the vault was abnormally high, as in oxycephaly. The most striking difference lay in the breadth of the head, which is reduced in oxycephaly but was increased in our patient. It was evident in this case, as has been proved by the pathologic report on the segment of bone removed from the coronal suture at the 1st stage of the operation (Fig. 2), that the deformity was due to the premature closure of the coronal suture, and that the skull in this child had expanded laterally (Fig. 3) and vertically (Fig. 4), because growth in the anterior and posterior planes was impossible. The question arose, therefore, as to how it would be possible to enlarge the capacity of the skull so that the brain might grow anteriorly and posteriorly. The two problems in this case were, (1) to relieve the increased intracranial pressure and thus preserve vision, and (2) to plan a second plastic procedure on the left side to correct further the abnormal shape of the head. None of the various operations advised for craniostenosis would permit the solution of this problem in this particular patient since all previous operations had been performed in cases of oxycephaly or scaphocephaly, but not for cases of decon-

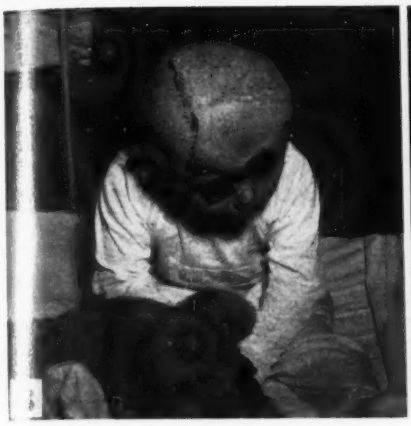

FIG. 3. The head is abnormally broad and short with marked diminution of the anteroposterior diameter.

FIG. 4. The head is flattened in the anterior and posterior plane, with the cranial wall abnormally high.

pensated brachycephaly. In view of the roentgenograms and the general configuration of the child's head it was thought that the operation should be based on a modification of King's procedure.

2nd Operation. A good-sized skin flap was turned down over the left temporoparietal region, proceeding well anteriorly to the closed coronal suture line and posteriorly terminating above the mastoid process. The area of the closed coronal suture, in addition to being thickened, showed that the suture was represented by a distinct white line through the bone just as on the right side. Burr holes were made around the periphery of the exposed skull about 3 to 5 cm. apart, and several other burr holes were made in the central part of the exposed skull. The holes were connected, using a Montenovesi forceps so that the area exposed was cut into 3 rectangular pieces of bone (Fig. 5). A small bridge of bone, holding the fragments together, was left until most

FIG. 5. Area of cranial vault cut into 3 rectangular pieces of bone.

of the operation was completed. The bone was thickened in the coronal suture area, as mentioned above, but particularly down toward the pterion where the bone made a much deeper projection inward than usual between the posterior end of the frontal and temporal lobe. A moderate amount of bone was removed in the temporal region as an additional helpful measure. This completed, the remaining bridges of bone were removed. The dura was not opened, owing to the fact that there was no papilledema at this time. Strips of gelfoam were placed between the fragments of bone. The skin flap was replaced and closed with two layers of interrupted silk sutures. It was noticed that there was some expansion of the skull wall, particularly over the frontal area, in comparison to the opposite side, at the

FIG. 6. Patient 3 weeks after 2nd operation, showing expansion of the skull wall, particularly over the frontal area. Compare with Fig. 4.

FIG. 7. Three months after 2nd operation. The shape of the head on the left side is practically normal.

completion of the operation. A dressing was applied with an outside protection of plaster shell.

The *postoperative course* was uneventful and the wound healed satisfactorily. The patient was discharged on Mar. 12, 1948 (Fig. 6), showing marked improvement both in physical condition and mental development, the physical improvement being pronounced as far as his gait was concerned.

Three months later the patient was seen at the clinic (Fig. 7); he showed constant pronounced improvement over his condition before he was operated on. He was able to walk freely and had begun to say a few words. The shape of the head on the left side was practically normal. The right decompression was full but not tense.

SUMMARY

A new classification of craniostenosis, based on anatomical and clinical facts, has been suggested.

An historical review of the evolution of surgical treatment of craniostenosis is discussed and analyzed.

A modified operation for the decompensated brachycephalic form is offered.

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SURGICAL EXPERIENCES WITH ACOUSTIC TUMORS

ALFREDO GIVRÉ, M.D., AND HERBERT OLIVECRONA, M.D.
Neurosurgical Clinic, Serafimerlasarettet, Stockholm, Sweden

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ACOUSTIC tumors rank third in the statistics of brain tumors operated on in our clinic. Out of a total of 3,265 brain tumors verified to Dec. 31, 1946 there were 304 unilateral acoustic tumors, or 9.3 per cent. The total number of neurinomas is somewhat higher as there were 6 cases of bilateral acoustic tumors and 4 cases of neurinomas arising from the 5th nerve.

In previous communications from this clinic,[2,3,4] the early and late results up to 1930 and 1939 were reported. In the present paper we shall review the entire material of acoustic tumors from December 1922, when the first case was verified, until the end of 1946. Although the material is fairly homogeneous in that nearly every patient has been examined by the senior author and all except 21 have been operated on by him, a number of new factors have necessarily come into play during this period of 24 years. The technical equipment has been enormously improved since the early twenties by the introduction of suction, the electrosurgical unit, and long-handled silver clip holders, to mention only a few of the many new devices that have been added to our armamentarium. New methods of anesthesia, particularly the intratracheal method, and the unilateral approach introduced by Dandy have relieved the surgeon of a good deal of his burden, lessened the operative shock, and made the operation less of a Marathon procedure than it used to be. These factors all contribute to simplify the technique of operation and have made possible the extensive use of more radical procedures than those in vogue 20 years ago. The realization that in those cases where some considerable portion of the tumor has been left behind few patients survive more than 3–4 years, and that few of the survivors regain their capacity for work, has led to the adoption of radical removal as the method of choice in acoustic tumors. It should not be forgotten, however, that the radical removal of an acoustic tumor carries considerably more risk than the more conservative methods carried out under present day conditions by surgeons of great experience, as shown by Cushing's[1] latest figures. It should be realized, therefore, that our statistics covering the last 24 years reflect the conflicting influences of improved equipment and technique and an increasingly radical attitude towards these tumors.

A sad fact remains to be noted—the diagnosis or rather the ability of the non-specialist to recognize the acoustic tumors in their early stage, has not improved much. We still receive most of our patients with acoustic tumors in a very late stage of development, nearly all of them with choked discs and not a few of them blind or with vision much reduced. Almost without exception a large tumor is found at operation and our records contain only a single instance of a very early tumor where the only symptoms were loss of

hearing and an enlarged porus, and this patient came under neurosurgical observation only because he was fortunate enough to have had a few vertiginous attacks suggesting Ménière's disease.

Among our 304 cases of acoustic tumors there was 1 in which operation was not performed. In 2 cases a suboccipital decompression was made. In 1 of these, where a reliable history could not be obtained, auditory perception was only slightly affected. Ventriculograms showed only a moderate degree of hydrocephalus. This patient died 3 weeks after the suboccipital decompression from pulmonary embolism. Autopsy showed an acoustic tumor on the side of the better ear. In the other case, where the tumor was thought to be too vascular to be attacked, death occurred 3 weeks later from pneumonia. In 1 case where the patient entered the clinic in a comatose condition only a subtemporal decompression was made. The patient died 3 weeks later from the effects of increased intracranial pressure.

The remaining 300 patients were all subjected to total or partial removal. In earlier communications the attempt was made to divide the partially removed tumors into 2 groups: the intracapsular enucleation and subtotal removal. However, on reviewing the case histories it was found that there was no significant difference in the subsequent course of the disease in the 2 groups, and since the division is an arbitrary one and apt to lead to confusion, especially when more than one operator is involved, the division of the incomplete removals into 2 groups was abandoned. All incomplete removals will therefore be treated as a single group regardless of the relative amount of tumor tissue removed. It is recognized, however, that this group is far from homogeneous. Especially in the early cases the intracapsular enucleation, which was the method of choice in those days, frequently amounted to little more than a suboccipital decompression, while with increasing experience relatively more tumor tissue was as a rule removed. It should also be pointed out that during the last 15 years an incomplete removal has rarely been deliberate; in most cases during this time radical removal was attempted, but had to be abandoned short of completion because of technical difficulties, complications occurring during operation and so forth. In many of these cases only a small part of the tumor was left behind. The unilateral approach used almost exclusively during the last 15 years also introduces another variable, as the decompression left after an incomplete removal probably is somewhat less effective than after a bilateral exposure. It is our impression, however, that the subsequent fate of the patient is less affected by the relative amount of tumor tissue removed and the size of the decompression than by the nature of the tumor and particularly its rate of growth.

Altogether 83 cases belong to the group in which the tumor was incompletely removed, while in the remaining 217 cases all visible tumor tissue was extirpated. This latter group may be further subdivided into 2 groups, one where the anatomical continuity of the facial nerve was preserved at the end of the operation (69 cases) and one where efforts in this direction were either unsuccessful or the nerve was deliberately sacrified (148 cases.)

TABLE 1

	1923–30	1931–39	1939–46	Total
Incomplete removal	32	43	8	83
Complete removal	0	71	77	148
Complete removal, facial nerve preserved	0	27	42	69
	32	141	127	300

The tendency towards more radical procedures in the treatment of acoustic tumors is clearly seen in Table 1. During the first period 1923–30, incomplete removal was used exclusively; in the period 1931–39, 30.5 per cent of the tumors were still incompletely removed; while during the last period, only 6.3 per cent were so treated. Among this last group there were 4 cystic tumors where for reasons to be discussed later an incomplete removal may be all that is necessary, and in 2 cases there was an urgent indication to save the facial nerve. In 2 cases only, the operation had to be abandoned short of completion because of technical difficulties or complications of some sort or other.

In Table 2 the operative mortality figures are presented. Taken at their face value these figures would seem to indicate that incomplete removal is the most dangerous method and that complete removal with saving of the facial nerve carries very much less risk than the other methods. In reality these conclusions are of course entirely wrong. The unfavourable showing of the incomplete removals depends largely upon the fact that during the early years, when the intracapsular method was used exclusively, the operator had very little experience and the mortality therefore naturally tended to be high. On the other hand, as experience grew, more radical methods were gradually adopted and the group of incomplete removals therefore tended to be composed of the most unfavourable cases, a considerable portion of them being cases where an attempted complete removal had to be abandoned because of technical difficulties, complications during operation and so forth. This group therefore is unduly weighted with unfavourable cases and the misfortunes of an inexperienced operator and there is little doubt that if the conservative method were to be rigorously adhered to, the immediate mortality could be reduced to an insignificant figure.

With regard to the mortality attending complete removal it is obvious

TABLE 2

	No. of Cases	Operative Deaths	Percentage
Incomplete removal	83	24	29
Complete removal, facial nerve preserved	69	7	10
Complete removal, facial nerve not saved	148	44	29.7

that the group where the facial nerve was saved is composed largely of the more favourable cases while the other group contains many more unfavourable cases. To this group also belong many cases where prolonged efforts to save the facial nerve had failed with resulting increase of the operative trauma.

The cause of death in the complete extirpations was: pulmonary complications (pneumonia, embolism, usually 1 to several weeks after operation), 5 cases; subdural hematoma, lesion of the basilar artery, injury to the vagus nerve, shock and hemorrhage, each 1 case. In all the remaining 41 cases death was caused by a postoperative clot in the tumor bed or hemorrhagic softening of the pons. Nearly always, both a clot in the angle and hemorrhagic softening of the pons were combined, but in most cases one of these was dominating. In about half of the cases the extracerebral clot was regarded as the most important cause of death; in the other half, hemorrhagic softening of the pons was extensive enough to be regarded as the cause of death and a small clot in the tumor bed was considered to be of minor importance.

Among the incomplete removals, pulmonary complications were responsible for 4 fatalities, and meningitis was the cause of death in 2 cases (1 cerebrospinal fluid fistula, 1 originating from opened mastoid cells). Shock and hemorrhage caused death in 3 cases, and 3 fatalities were due to postoperative pressure associated with backward dislocation of the brain stem because of incomplete tumor removal. In the remaining 12 cases death was caused by a postoperative clot within, and usually also outside, the tumor capsule, nearly always combined with hemorrhagic softening of the pons.

The reasons for this predominance of postoperative clots as the cause of death is, of course, that even a small clot, which would be of little consequence in a large cavity in one of the cerebral hemispheres, is very dangerous in the angle, especially when associated with more or less swelling of the adjacent structures. Hemorrhagic softening of the pons may sometimes be caused directly by the operative trauma but, in our opinion, more often by the obstruction by clips or coagulation of the petrous vein and other veins on the lateral side of the pons. The reason for this belief is that softening of the pons may often be seen even when removal of the tumor has been easy with practically no direct damage to brain stem during operation.*

Among the 59 survivors of the incomplete operation 5 have been lost track of, in most cases because the patients lived in central Europe. Twenty are known to be dead, the proven or probable cause of death in every instance being recurrence of the tumor. The average survival period in these cases has been a little more than 3 years after operation. In 9 cases the patients came back to the clinic with recurrence. In all these cases the tumor was completely removed, with 4 fatalities, 1 from pulmonary embolism, the cause of death in the remaining 3 cases being softening of the pons or a postoper-

* After completion of this paper the article by W. J. Atkinson (*J. Neurol., Neurosurg. Psychiat.*, 1949, n.s. *12:* 137–151) on the variations of the anterior inferior cerebellar artery came to our attention. This important paper gives a very good explanation of the hemorrhagic softening of the lateral part of the pons, so frequently observed after extirpation of acoustic tumors.

ative clot. In the cases terminating fatally the 2nd operation was performed on the average 5½ years after the 1st. In the surviving cases the average time between operations was 4½ years.

Altogether there were therefore among the 59 survivors 29 recurrences, or 50 per cent, the recurrence leading to death or to a 2nd operation between 3–4 years after the 1st operation. The mortality rate with and without secondary operation was 40 per cent of those surviving the 1st operation. The total mortality for incompletely removed acoustic tumors therefore is exceedingly high. Within 3–4 years 60 per cent of those so treated are dead either from the immediate effects of the operation or from recurrence. Even if this figure could be somewhat reduced by a lower primary mortality, the late mortality from recurrence would probably remain unchanged. This must be considered a strong argument in favour of radical removal of acoustic tumors.

The functional results in the survivors showing later recurrence after the incomplete operation are equally depressing. In none of the 29 recurrent cases was full earning capacity ever regained excepting those cases where the tumor was later successfully removed. Among these 5 cases, 4 regained full earning capacity, and 1 was much improved but died some years later from the effects of a second neurinoma in the brachial plexus. With 1 or 2 exceptions the other patients with recurrence were all invalids, although some of them were able to do light work at least for some time before recurrent tumor symptoms became manifest. Cerebellar incoordination was the dominant cause of inability to work in nearly all cases, although poor vision, headache, mental disturbance and so on were contributory in some instances.

If we now turn to those who are known to be still alive it is rather surprising to find that among these 25 patients surviving for an average period of 13 years, 12 consider themselves completely well with full earning capacity, 9 are in a fairly good condition with earning capacity ranging between 50–75 per cent and only 4 are complete invalids, 2 because of blindness and 2 because of cerebellar disturbances. Among those who are well, there are 5 cystic tumors, where nothing was done except evacuating the cyst and cauterizing its walls with Zenker's fluid. These 5 patients have been under observation from 5 to 20 years, the average observation time being 12 years. Four with solid tumors have been followed for an average of 19½ years. Since the technical procedure has been the same as in those cases recurring within a few years it is hardly possible to escape the conclusion that some acoustic tumors do not continue to grow after incomplete removal or at least grow so imperceptibly slowly that life and function are not endangered for many years or even decades. Unfortunately there is no way except in the case of the cystic tumors to decide at the time of operation how the tumor is going to behave in the future. In the case of cystic tumors, however, there is good reason to be conservative and even if some of these do recur, the time until recurrent symptoms manifest themselves is usually a long one and quite frequently 10–15 years or more elapse without any symptoms of recurrence.

After complete removal of the tumor there were 166 patients surviving the operation. Eighteen of these have not been heard from for some years and as most of them were Jews from Central Europe they are presumably dead. Nine patients are known to be dead, 1 from pneumonia 2 months after leaving the hospital, the remaining cases between 1–10 years later, all from intercurrent disease. In 1 case which came to autopsy a recurrent tumor the size of a pea was found. Otherwise no recurrences have been observed.

FIG. 1. N. C. 225/40. Woman, aged 48 years. Right-sided acoustic tumor completely removed 1940. (A) Picture taken 7 days after operation. (B) 7 years after operation.

There remain therefore 139 patients who are still alive and whose subsequent fate is known. Among these, 63 or about 50 per cent are well with full or nearly full capacity for work; 54 are fairly well but their earning capacity is reduced, usually to about 50–75 per cent. There are 22 who are complete invalids, 7 of them because of blindness which was present already before operation. In the remaining cases cerebellar incoordination and general weakness, sometimes also associated with old age, were the chief causes of invalidism. The average observation time of the surviving cases is 7 years. In some of the recent patients who have been followed for only a year or so, and who have been classified as complete invalids, some recovery of function may be expected but probably none of them will ever regain full earning capacity.

Of the 62 survivors where the anatomical continuity of the facial nerve was preserved, 6 have not been heard from. Among the remaining 56, there was complete or nearly complete recovery of function in 19, partial recovery

in 20, and no recovery in 17. In two-thirds of the cases, where the facial nerve has been saved, moderate to good recovery may be expected, while in one third a complete facial paralysis persists. It should be noted that in every case the porus has been coagulated to destroy all remnants of tumor tissue. This naturally leads to complete loss of function immediately after operation and in a number of cases there is probably too much scar tissue for regeneration to be possible.

It has already been noted that the operative mortality in the group where the facial nerve has been saved is much lower than in the other groups. The results with regard to earning capacity also are considerably better. There are only 4 complete invalids, 2 because of amaurosis, among the 56 patients

FIG. 2. N. C. 575/46. Woman, aged 37 years. Right-sided acoustic tumor completely removed 1946. Pictures taken 1 year after operation.

who could be followed, as compared with 18 (5 cases of blindness) among the 139 cases where the facial nerve could not be saved. The time of rehabilitation is also considerably shorter in this more favourable group, where the tumor as a rule is smaller and more easily handled.

A comparison between the final results in the incomplete and the complete removals shows, that in the former group 25 per cent are reasonably well and able to work after an average observation time of 13 years, while among the complete removals 55 per cent are well and working after an average observation time of 7 years. There is no mortality from recurrence where the tumor has been completely removed, while among the incomplete removals 40 per cent died within 3–4 years from recurrence or secondary operations.

A final word should be said about the influence upon the mortality rate of attempts to save the facial nerve. In our opinion the careful dissection of the facial nerve makes the operation infinitely more difficult, the operation is considerably prolonged thereby, and although the increase in the operative risk cannot be stated in figures, there is in our opinion no doubt that the danger of the operation is to some extent increased by efforts to save the

facial nerve. Although it is eminently desirable to save the facial nerve, this principle should not be carried too far and the cases should be carefully selected with due consideration to the age, sex and occupation of the patient. A serious effort to save the nerve should be made only when the anatomical conditions in the field of operation are reasonably favourable.

The general conclusion from our present experience with acoustic tumors is that they should be completely removed unless the tumor is cystic or there is a very urgent indication to save the facial nerve, and the anatomical

FIG. 3. N. C. 199/40. Woman, aged 21 years. Left-sided acoustic tumor completely removed 1940. (A) Picture taken 6 days after operation. (B and C) 4 years after operation.

conditions in the operative field are not quite favourable for saving the nerve.

TECHNIQUE OF OPERATION

We prefer local anaesthesia, especially because the function of the facial nerve can be watched during the operation. Pain may occur, particularly when the tumor is mobilized from the glossopharyngeal nerve and also as the tumor is dissected away from the trigeminal root. Pain from the glossopharyngeal nerve can nearly always be prevented by placing a pledget of cotton, wet in 5 per cent novocain solution, on the nerve as it leaves the brain stem. Pain from the trigeminal nerve may occur both during the intracapsular part of the operation, though this is usually slight, and particularly as the capsule is pulled away from the root and the brain stem. This pain is difficult to prevent as the 5th root is not accessible until the capsule has been mobilized from the brain stem to a considerable extent. A few drops of novocain may then be injected into the root as it emerges from the pons.

It must be admitted, however, that the operation under local anaesthesia is not as free from discomfort for the patient as one would wish. The alternative to local anaesthesia is intubation with ether and oxygen. It is important, however, that the anaesthetist be very experienced, as even comparatively slight obstructions of the airway might cause considerable venous stasis and swelling of the cerebellum. As it is necessary to have all the room possible in this narrow field of operation, even

slight or moderate degrees of venous stasis might greatly increase the difficulties and hazards of operation. Needless to say, shoulder supports are absolutely essential especially if general anaesthesia is used. The face-down position with complete muscular relaxation induced by general anaesthesia, may during a prolonged operation place such a load on the heart, especially in elderly patients, that circulatory failure may occur.

In our clinic the face-down position is always used. It is important to place the patient with the neck well flexed and the head rotated somewhat away from the lesion. The head is fixed in this position with strips of adhesive tape. The table should be so constructed that it may be tilted around its long axle. During removal of the tumor the table is tilted this way, whereby a good exposure of the cerebellopontine angle is assured. The unilateral exposure proposed by Dandy is always used and the bone over the cerebellar hemisphere on the side of the lesion is removed. It is important to remove enough bone to expose the sigmoid sinus. Sometimes the mastoid cells extend behind the sinus, but unless there is a history of mastoid infection, the cells are opened to allow sufficient removal of bone. The opened cells are filled with penicillin solution and plugged with wax. After tapping the lateral ventricle, the dura is opened in stellate fashion. After retracting the cerebellum, the tumor is usually easy to expose and after incising the arachnoid covering the tumor, as much as possible on the surface of the tumor is exposed by pushing away the cerebellar cortex covering the neoplasm. After coagulating all superficial blood vessels visible in the capsule, the tumor is incised and as much as possible of the contents evacuated with a spoon and sucker, as in an ordinary intracapsular enucleation. During this procedure and until the end of the operation, a special nurse is watching the face of the patient. If the intracapsular enucleation is carried too close to the facial nerve, twitchings occur in the face, indicating that further progress of the enucleation in this direction should be stopped. From time to time the patient is asked to move the face on the affected side.

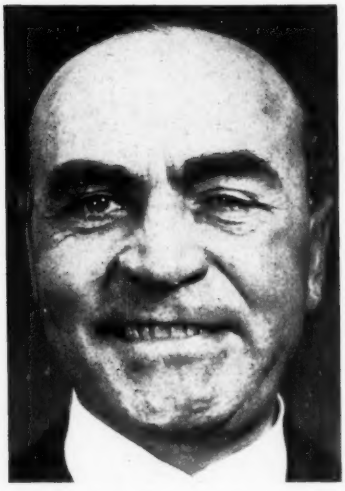

FIG. 4. N. C. 270/42. Man, aged 46 years. Left-sided acoustic tumor completely removed 1942. Picture taken 3 years after operation.

When the intracapsular enucleation has been carried sufficiently far, hemorrhage from the cavity is stopped by temporary packing with cotton strips, wet in 3 per cent hydrogen peroxide solution.

When a reasonable degree of hemostasis has been obtained, a de Martel automatic brain spatula is inserted, retracting the cerebellum from the lower pole of the tumor. The lower pole is now mobilized by retracting it upwards with a brain spatula. It is usually easy to separate the lower pole from the 9th 10th, and 11th nerves which are usually elongated and pushed downwards by the tumor. Frequently there are 1 or 2 fairly large branches from the vertebral artery coming from below and running up to the ventral part of the tumor or between the tumor and the brain

stem. These arteries are doubly clipped and divided. A search is then made for the facial nerve, which always runs on the ventral side of the tumor. Usually, the nerve is most easily seen as it runs as a flattened band on the lateral surface of the brain stem. After the nerve has been seen, its identity is confirmed by faradic stimulation. One should then try to follow the nerve in the lateral direction as far as possible. This is rarely possible to any great extent, because the nerve is always pushed away in the cranial direction and usually can be followed for only about 2 cm. from the brain stem. The nerve is usually somewhat adherent to the capsule of the tumor and should be freed by careful blunt dissection. If the tumor is small, it is sometimes possible to follow the nerve all the way to the porus, but in most cases the facial

FIG. 5. N. C. 661/39. Woman, aged 38 years. Right-sided acoustic tumor completely removed 1939. (A) Picture taken 8 days after operation. (B) 8 years after operation.

nerve crosses over the trigeminal nerve about 15 mm. from the porus to enter the porus at its anterior margin, about mid-way between the superior and inferior quadrant.

After the facial nerve has been dissected free as far as possible, the upper pole of the tumor, which is usually wedged in between the brain stem and the margin of the tentorium, is mobilized, if possible, so far that the trigeminal nerve can be seen at its emergence from the pons. If necessary, a few drops of novocain are injected into the nerve.

The next step is to mobilize the tumor from its attachment to the porus. There is always a projection of neoplastic tissue into the porus and the capsule of the tumor is always firmly adherent to the margins of the porus. A knife must be used to separate the tumor from the margins of the porus. Before doing this, it might be advisable to coagulate the petrous vein which lies on the superior surface of the tumor. It may be quite large and may be troublesome, unless coagulated before the tumor is separated from the tentorium.

The dissection of the tumor away from the porus must be done very carefully

to avoid injury to the facial nerve. As already pointed out, the usual point of entry of the nerve into the porus is in the anterior-inferior quadrant. Frequently, the nerve is separated from the capsule by a thin sheath of arachnoid, and, if existing, this sheath of arachnoid should be carefully preserved. As soon as the nerve is seen, it is identified by faradic stimulation. The neoplastic tissue present in the porus can usually be removed by blunt dissection without injury to the nerve. Nearly always there is troublesome bleeding from the porus which should be temporarily stopped by packing the porus with a small cotton pledget. When the facial nerve has been identified at its point of entry into the porus, it is carefully dissected away from the capsule. Usually the adhesions are quite firm between the lateral part of the nerve and tumor, and sometimes it is necessary to use a knife to separate these adhesions. If possible, the nerve should be dissected free from the capsule of the tumor in its entire course from the brain stem to the porus. When this has been accomplished, the removal of the tumor is a comparatively simple matter, provided the capsule is sufficiently tough to allow some pull to be exerted on it. If this is the case, the part of the capsule that has been attached to the ventral part of the porus is grasped with a Brüning nasal clamp and the capsule pulled away from the trigeminal and facial nerves. The medial part of the capsule is usually deeply embedded in the pons and several small arteries may be encountered, running from the lateral surface of the pons to the tumor. These are clipped and divided as they are seen and the pull on the tumor in a posterior-medial direction continued until the tumor is delivered from its bed in the cerebellopontine angle. A few veins on the lateral surface of the pons are usually torn in the process, but hemorrhage from this source usually stops by itself after temporary packing with strips of cotton, wet in hydrogen peroxide. The function of the facial nerve is now tested—in most cases the nerve has been so traumatized that voluntary function is lost, but response to faradic stimulation is usually retained, unless the nerve has been torn. The porus is now examined for remaining fragments of tumor. It is always advisable to coagulate thoroughly the porus both to destroy remaining tumor cells and to control hemorrhage, which may be quite troublesome from small arteries perforating the bone in the porus. During this process, any remaining function of the facial nerve is of course lost, but provided the anatomical continuity of the nerve is preserved, regeneration will take place in 2/3 of the cases. The pack in the angle is now removed and the lateral surface of the pons inspected for bleeding points. Arterial bleeding should be stopped by clipping the artery or by coagulation. One should be very careful, however, in coagulating blood vessels on the lateral surface of the pons. Venous oozing which has not stopped by itself or by temporary packing is best controlled by small pieces of gel foam pressed against the bleeding point for a short while.

After all hemorrhage is under control, the cavity is flushed out with 3 per cent hydrogen peroxide solution and normal saline. It can not be too strongly emphasized that control of hemorrhage must be absolute, as a fatal outcome of an operation for acoustic tumor is nearly always due to postoperative hemorrhage. The dura is carefully closed as watertight as possible.

SUMMARY

The immediate and late results in 300 operations for acoustic tumors are presented. In 83 cases the tumor was incompletely removed. In the remaining 217 cases all visible tumor tissue was extirpated. In 69 cases, or about 30 per cent, the anatomical continuity of the facial nerve was pre-

served. The mortality in the incompletely removed tumors was 29 per cent. In the completely removed tumors the mortality was 23.5 per cent. In two-thirds of the cases, where the facial nerve was saved, good or satisfactory regeneration occurred.

Among the survivors of the incomplete operations, recurrence of the tumor was observed in 50 per cent, the recurrence leading to death or to a 2nd operation 3–4 years after the 1st operation. The mortality rate with and without a 2nd operation therefore was 40 per cent of those surviving the 1st operation.

Among the completely removed tumors no recurrences were observed. The technique of operation is described.

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REMEMBRANCE OF THINGS PAST*

DANIEL C. ELKIN, M.D.
Emory University, Georgia

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DR. CRAIG, MEMBERS OF THE CUSHING SOCIETY AND GUESTS:

I AM most appreciative for the honor of the invitation that accounts for my presence here tonight. Individually, and socially, I have long known many of the members of this Society, but never, in my fondest dreams, could I have hoped to be your collective guest. Although having suffered from long exposure to neurosurgery, I have never been so emboldened as to practice the art. Perhaps that is why you invited me.

My appreciation is commingled with a sense of humility, I may say inferiority, and you must bear with me, for I admit to being out of character and out of place. I find myself much in the position of a mountaineer who went fishing at night. His friend rowed the boat in the bow, while he sat in the stern armed with a fishing pole, a bucket of minnows, a can of worms, and a jug of moonshine. He would cast his line and then take a drink from the jug: a perfect synthesis of two of the most delightful avocations of which I am aware. After this had gone on for some time, he was startled by an object in the water, and he said to his friend, "What's that?"

"Why that," said the friend, "is the moon."

"The moon! And what in the hell am I doing up there?"

Dr. Cushing's work and his life have been so covered in prose and verse, in the main by members of this Society, that little is left to be said. Since all the facets of this many-sided man have been so admirably dealt with in John Fulton's definitive *Life*, anything that I may say is by way of supererogation.The field of fact having been so amply covered, there is left to me only the expression of personal recollections; perhaps some fiction.

> The sense of the world is short,
> Long and various the report. . . .

The difficulty of speaking of Harvey Cushing is that he has become a thing apart, set aside as an immortal, which he certainly *is*, and we are apt to forget not so much the things that made him great, but the things that made him mortal, which he certainly *was*.

Surely if ever the imagination is allowed to riot, it has the privilege in our present environment. For Harvey Cushing remained to the last, a *professional* Yale man, a not unusual accomplishment, particularly since m

* Presented June 3, 1949 at the meeting of the Harvey Cushing Society, held at Yale University School of Medicine, New Haven, Connecticut, in commemoration of Doctor Cushing's eightieth birthday

his days at New Haven, Yale held a dominant place in athletics, and he was always "a part of all he met." As a member of the baseball team, his interest in Yale sports never waned, and like many another Old Blue, he could not refrain from occasional reference to the "good old days," and deprecation of the poor coaching which he assumed (correctly) to be the cause of Yale's descendency.

On one occasion shortly before his death I jokingly suggested to him that there was a movement on foot to bring Walter Camp back as the Yale coach. I immediately knew that I had touched a sore point. He at once realized that I was speaking irreverently of a Yale god, and he furiously turned on me with the remark: "Well, he would at least teach them how to tackle."

Like most graduate students he never considered himself an alumnus of Harvard. His return here was in a way a college reunion. *Brekekekex* was always sounding in his ears; the crack of bat on ball, the old friendships of field and fence and classroom . . . so it is most appropriate that we meet tonight beneath the elms.

For those of you who did not know him, perhaps a personal recollection of his appearance might be in order. He had a long neck, which Sargent, according to his custom, caricatured with his crayon. His brow was wide and high, that of a thinker, but of particular serenity. His walk had the easy swing of the born athlete, but it was the head that attracted attention and disarmed you of the size of the body, for he was short and small.

He always reminded me of a jockey grown a little too old to ride, but with the figure and force and the strong hands, the vim and vitality to be a trainer, and, if the occasion demanded, to saddle a horse for himself.

His mouth, as I recollect it, was his best feature; the chin sharp but not imperious, and the nose long and big, but all were blended well—a man of action—but at the same time expressing poise and equanimity. There was both power and thoughtful speculation blended in that face, a face that was never doubtful of its power, but one that was not conceited, for above all things he hated humbug in any form.

He dressed well, a little nattily, usually grey tweeds of an ill-fitting, English manner, never gaudy save for large ties knit by Mrs. Cushing, which he chose to wear when Windsor was only a prince.

Perhaps it is old-fashioned to think of an inheritance like this, but in him was commingled the inherent sanity of New England Puritanism crossed with the pioneering spirit of the West, and all leveled out to produce what Galsworthy called "the soul of balance."

His outstanding characteristic, save his ambition, was his capacity for work—coextensive terms, to be sure. He never allowed the duties of administration to interfere with the development of neurosurgery *and* of Harvey Cushing. He served on few committees, and I recall no instance of a formal meeting of his staff, save on the occasion of Gil Horrax's marriage, which was used as an opportunity for pointing out the marring and disastrous

effects of the connubial state on the mental development of a surgeon.

His prodigious capacity for work was a discipline which apparently began in his undergraduate years, or perhaps before, but which never reached its peak until the Hopkins days. He fitted his entire life into this whole program. He cared nothing for the theatre and less for music. His avocations were travel, book browsing and collecting, his social and literary societies—notably The Club, The Saturday and The Elizabethan. Above all, he loved good talk.

The absolute adherence to the principle of geographic, full-time concentration of his work accounted in no small part for the tremendous accomplishments. His trips away from home, from first to last, were for the purpose of his continuing education, for meetings, for lectures, the receipt of honorary degrees, and rarely if ever for consultations, and never for the purpose of operating.

In the preparation of his papers, as in the operating room, he was a perfectionist, and he worked over them to the last minute before presentation or publication. He had a *penchant* for a foreign or classic word or phrase, placed in his writings with great care, and in such a way that the context could be easily understood, for he was not pedantic. Every patient that he saw, even from his interne days, was considered a possible case report, and no small detail, even to the point of his own sketches, was omitted. Writing apparently did not come easy to him in the early days, and one can follow the evolution of this art which he brought to such perfection, by reading his papers chronologically.

He demanded from others, but rarely received it, the same discipline. I recall that when Sir Harold Stiles was Professor of Surgery *pro tempore*, Francis Newton and I used this distinguished scholar as an excuse to attend a baseball game. As usual, Dr. Cushing caught us on our return to the hospital, and in Sir Harold's presence reminded us that Sir Harold was there to teach students and not to be instructed in the intricacies of American sports. That night at dinner, however, he spent a greater part of the evening explaining to Sir Harold the finer points of the game.

In relation to money matters he was no different from the ordinary run-of-the-mine doctor. His services were sought wide and far, by rich and poor, who were treated accordingly. He had a keen sense of value, certainly of his own, and from the wealthy he received good, but never exorbitant, fees. He was practical about money matters and rarely did he operate without previously discussing, in a frank manner, his fee, with the patient or with a member of the family.

Time and again he was asked to operate away from home, and at any fee he might request. This was always refused for the obvious reason that such excursions would interfere with the running of his own clinic.

While he never threw money away, he could be generous in the extreme, particularly in supplying small needs for house officers, for travel expenses and reprints, for which there was no reimbursement from his own depart-

ment. In the collection of his books which he planned to give away, he was a profligate spender, even in the years after he had suffered severe financial reverses.

He was paradoxical in the extreme, both in his actions and in his statements, some of which were made for the purpose of startling his listeners. He would talk seriously of the value of detailed knowledge of anatomy as essential to surgery, and conversely, he was known to have said to one of his house officers, "What's the use of anatomy? When you come to a blood vessel, you cut it and tie it. That's all there is to anatomy."

He would talk mightily, and in fact write papers about the glory of the general practitioner, but no man in his day was more given to specialization. He had tremendous admiration for the benefits of group practice, but certainly never practised it himself. He was a great believer of full-time practice, but only in its geographic sense, and, almost singlehanded, stood out against the General Education Board and Harvard University as a protagonist for individual practice in its entirety.

The striking thing about his personality was that apparently he had a set plan for his life, and nothing, so far as I know, was allowed to interfere with it. Other men have planned likewise, usually unsuccessfully because in the end they brought down upon themselves the combined antagonism of those whom they had used in their ambitious ascent to power, for those who follow that course often do so at the expense of others. It is not remarkable in his achievement that Dr. Cushing stepped on a few toes; the astonishing thing is that he did not mutilate more. On the other hand, in the accomplishment of his great ambitions there were thousands who benefited by his knowledge and skill, and hundreds whom he advanced to positions of greatness and trust.

Dr. Cushing was certainly sure of his position in history, as many men have been, but rarely did he give any indication of it. His literary executor became well aware of it.

Like many people who attain great success, he could do big things in a big way and small things in a small way. He was human. He could bear grudges about small matters, bear them in fact until the mellower years of his life. But at the same time he had a tremendous capacity for loyalty which could overlook shortcomings and minor vices of his friends and pupils.

He was affable, and when he wanted to, could charm anyone, particularly visitors, or his juniors. He had a knack of putting them at their ease, and the greater knack of getting rid of them without injuring their feelings. He brought to the "bum's rush" the manners of the parlour. In the great Brigham days, he had visitors by the hundreds from the world over. They rarely interfered with his work. He would greet them with effusion, place them in the operating room (where they could not leave), take them to lunch and around the wards, and, particularly if they were foreigners, ask questions about their chiefs-of-clinic which they were sure to take home.

A favorite trick of getting rid of a visitor was to ask where he was stop-

ping. If Gus were at hand, Dr. Cushing would escort the visitor out the side door to the car and send him off to his hotel. Otherwise, he would draw a map which included his office, Huntington Avenue, The Fenway, and the Copley Plaza, and usher him to the door, and off would go the visitor, with a gracious smile, a pat on the back, and a trophy in the form of a map to carry home to his chief as proof of his visit.

He could be rude, too, often to his own later embarrassment. This was probably the result of his abstract thinking, innumerable worries, his practice, and a thousand things that enter into a man's life.

Although brusque, nagging, at times impatient, and a hard taskmaster, he never, to my knowledge, embarrassed an operative assistant, in spite of many stories to the contrary. His favorite admonitions, "Keep your eye on the ball," and "Don't watch the clock," were usually *sotto voce*. Occasionally he would take great pains to answer a question from the stand or from a new assistant, but as a rule the question went unnoticed. He taught by precept and not by spoon.

To illustrate his renowned abstraction, usually interpreted as rudeness— I will not call it rudeness— a new assistant resident during my Brigham days related this story:

"About two weeks ago," he said, "I had been holding a retractor for about four hours, and I noticed that The Chief had left a small cotton sponge in the corner of the wound, beneath the dura. He was beginning to close, and with some fear and trembling I said, 'Pardon me, Dr. Cushing, I think there is a sponge in the corner of the wound,' and pointed to it with an instrument.

"He lifted the dura, picked out the sponge, threw it on the floor and testily said, 'I knew it was there all the time.'

"Well," continued the assistant, "I knew better. I felt I had been mistreated and I thought he ought to have thanked me. So I bided my time.

"This morning," he said, "during a cerebellar operation, I saw him place another sponge just where the other one had been. I watched it for an hour and he hadn't touched it. Another hour went by and I became fascinated by the game. 'At last I've got you, old boy, and you won't insult me again!'

"Three more hours passed. I let him close down to the skin—an hour's job. Then I said again: 'Pardon me, Dr. Cushing, I think you left a sponge in the corner of the wound.'

"'Why didn't you say it was there sooner,' he said.

"'I thought you knew it was there all the time.' I replied.

"He said not another word, but carefully cut the sutures and explored the wound.

"'Now,' said Dr. Cushing, 'Where is the sponge?'

"I said, pointing to the edge of the dura, 'Under there, Sir.'

"And what do you suppose he found?"

"A sponge," I said.

"No. Not a damn thing."

Dr. Cushing had a fine sense of humor, but again most individualistic

Both his jokes and his sayings frequently had some literary or historical bearing. He liked the pun, but usually laid this form of wit to the door of another. He would go to great ends in order to perpetrate a practical joke, as in the well-known incident of Chevalier Jackson. He was a born actor and could dramatize a small incident and vivify it by phrase and gesture. He well knew that the art of story telling frequently hinged on one's ability to satirize himself. He loved above all things to tell how Gus, the major-domo of the Cushing household, or Adolph, the czar of the Brigham, could put him in his place. He thoroughly enjoyed being taken off in the amateur theatricals of the Brigham staff, in fact, would have resented it had he not been.

His wit sometimes took the form of pulling the leg. For instance, after a long series of Yale defeats, the football team finally defeated Harvard at Cambridge in 1923. This was the beginning, if I am not mistaken, of the many assaults on the goal posts by the victorious students. The next morning, Dr. Cushing wrote a letter to the Harvard Athletic Association, apologizing for the incident, and asked them to submit a bill for the damage that had been done, and which he, as a Yale alumnus, would be glad to pay. However, he added this postscript: "Please let me have the wholesale price on the goal posts, because we expect to do this for a good many years in the future."

He had a capacity for making and holding friendships, for the most part those of college days or early medical associations. He had a temper which was apt to take fire and burst out, but it was just as quick to subside. Those who felt his ire were apt later to find his arm stuck through theirs or over the shoulder, with a disarming question about some purely personal matter which meant in a word, "I am sorry," or "All is forgiven," or just, "Let's forget it."

He was always thoughtful, or rather, I should say, thinking, particularly at medical meetings, or at the presentations by his own staff, which was often mistaken for dourness.

He had an inherent sense of justice but it was curiously tempered with his belief that most men were born with equal opportunity, that pampering was bad for the soul, and that failure was probably a man's own fault, and due to laziness. For this reason he could never quite bring himself to believe in the New Deal, and in this connection he once said to me: "*Kate* thinks he's a smart fellow."

You well know that his reputation was made in a new and unknown institution of 250 beds. Within 10 years, The Brigham was the best known in the country, which is the finest example that I know that individuals make an institution.

His teaching, as I have said, was wholly by precept. He never had a staff meeting as such, but no individual left his office or his home without some germ being planted about his feeling regarding the working of the hospital, or an admonition for some shortcoming.

He was a hard taskmaster, and rarely was there praise; the latter was

reserved for future years, and this frequently came in the discussion of a paper at a medical meeting, or a note regarding a publication. These were not always in praise, but at times contained a fatherly warning. A few months after I left the Brigham, I had in mind the publication of an article on a historical subject, and wrote him for advice. This he gladly gave, but added: "Beware of too much historical writing at this stage. It is not understood by many folks. Most people are apt to think that historical study is incompatible with good surgery. What you will need is one or two good surgical papers every year. They will have to be better, I may add, than that skit you sent me about cirsoid aneurysm."

My own association with Dr. Cushing was a fortunate one, and varied, by pure chance, from that of the other members of his house staff. I had met him first by happy circumstance. I was a senior in college, a member of his Society. He was an alumnus who expressed an interest, and took it, in my proposal to study medicine. It was the beginning of a long and intimate friendship, in a sense fraternal, and enhanced by the fact that the love and admiration in which I held him was never stultified by awe or fear.

I could cite, were I less emotional, innumerable instances of his kindness, great and small, to me and to members of my family, which were ample evidence of a great heart and soul, so often forgotten and overlooked in the busy life of an eminent man.

And where does this restless spirit roam tonight? New England and The Western Reserve with his forebears and the boy he came to love so well? Harvard and Hopkins with their promise of the greater days to come? Oxford and Osler? The Brigham which saw the full fruit? Here surely it rests tonight among those things he cherished most in life—his friends. Here at last the serenity that comes with appreciation of accomplishment. Here his books. Here the preservation of the material evidence of his greatness. And here with this company of whom he wrote:

"I am very proud of you all. And that I should have been immortalized by having you use my name is a source of pride and gratification."

CASE REPORTS AND TECHNICAL NOTES

INTRACRANIAL CERUMINOUS ADENOMA

LOUIS BERLIN, M.D.*
Neuropathology Laboratory of the Neuropsychiatric Institute, University Hospital, Ann Arbor, Michigan

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Of the intracranial tumor masses responsible for the syndrome of progressive unilateral deafness, the best known is the acoustic neurinoma. However, other tumors, either primary or metastatic, may occur in the same region and give rise to a similar syndrome. The most common primary tumors to be differentiated from an acoustic neurinoma are the meningiomas of the cerebellopontine angle, gliomas of the pons, and cholesteatomas.[2,7,12] The metastatic tumors that invade that area are the carcinomas and abscesses, especially those arising from the external and middle ear,[6,7] lymphoepitheliomas, angiomas of the middle ear,[5] carotid body tumors, malignant melanomas, and sarcomas of the temporal bone.[7] To this list of tumors involving the 8th nerve should now be added the ceruminous adenoma.

The ceruminous adenoma is a rare tumor that usually arises from the ceruminous glands of the external ear. It is made up of tubules or alveoli of various dimensions which may become distended into cystic cavities. These may then present the appearance of cystadenomata. The epithelium of the tumor is generally composed of two layers of cells, an inner layer of cuboidal to cylindrical cells, and an outer layer of flat cells. The epithelium may be thrown into papillary folds within the cystic areas. The tumor is surrounded by a fibrous capsule and there are also numerous connective-tissue septa between the glandular elements.

Since the first description of this tumor by Haug[8] in 1894, a total of 6 cases have been described.[1,4,9,10,11,13] In every instance the tumor was present only in the external ear. It is the purpose of this paper to report for the first time the intracranial occurrence of a ceruminous adenoma and discuss the clinical and pathologic diagnostic problems which this presents.

CASE REPORT

J. S. (631642), a 26-year-old white farmer, was admitted to the University Hospital on Feb. 16, 1948, because of deafness in the left ear, and left facial paralysis. The first symptoms were noted 9 years previously, when he suddenly discovered during a telephone conversation that he was deaf in his left ear. One and a half years prior to admission there was noted also some flattening of the left side of the face and this progressed to a complete left facial paresis 8 months before admission. There was no associated headache, dizziness, tinnitus, ataxia or weakness. The rest of the past history and family history was non-contributory.

Examination. There was an almost complete, peripheral type of facial paresis which permitted the slow closure of the eyelid, and a complete nerve deafness on the left. The left tympanic membrane was hyperemic and the light reflex was absent. There was no other evidence of impairment of sensory or motor function anywhere, no ataxia, cerebellar dysfunction or increased intracranial pressure.

X-rays of the skull showed a destructive process involving the petrous portion of the left

* Now at Winter Veterans Administration Hospital, Topeka, Kansas.

temporal bone (Fig. 1) and obscuring the internal auditory meatus. There was a minimal irregular calcific density within the region of the lesion. This was interpreted as representing a malignant neoplasm arising at the base of the skull and invading the cerebellopontine angle. It was not considered to be the x-ray appearance of an acoustic neurinoma.

Operation. On Feb. 20, 1948, a left suboccipital craniotomy was performed by Dr. Max M. Peet. After the dura was opened, a tough gray circumscribed tumor was encountered which extended from the petrous ridge of the left temporal bone posteriorly to the tentorium cerebelli and superiorly for a small, but undetermined distance above the tentorium and inferiorly down to the pons. The tumor was seen to displace the left hemisphere of the cere-

FIG. 1. Destruction of the petrous portion of the left temporal bone by the tumor. Note also the calcific density within the tumor.

bellum downward and to the right. It was located entirely extradurally. Exploration of neighboring subdural and subarachnoid spaces failed to reveal any extension of the tumor. Because of the size of the mass, it was necessary to perforate it and thereby decompress it in order to facilitate removal. It was estimated that the tumor weighed 50 grams.

Postoperative Course. The immediate course was marked by some difficulty in swallowing and paralysis of the uvula. Bronchoscopy had to be performed on the 3rd postoperative day in order to aspirate the accumulated secretions. However, by the 8th day the patient was able to swallow and the weakness of the uvula had diminished. At the time of his discharge, on Mar. 8, 1948, there were residual 6th nerve weakness, complete facial paresis, nerve deafness, and partial paralysis of the uvula, all on the left side. The gag reflex was present and the patient was able to swallow liquids and soft foods. There was no other sensory or motor impairment and no ataxia.

Pathologic Report. The specimen consisted of a firm, fibrous, spherical mass, $2\frac{1}{2}$ cm. in diameter, with numerous irregular knobs on the surface. Upon section the tumor was seen to contain numerous cystic areas filled with a brown, hemorrhagic fluid undoubtedly introduced during the operation. The tumor was basically an adenoma, composed of tubules which were often distended into cystic cavities. The epithelium of the tubules was composed of two layers

of cells, an inner layer of low cuboidal to columnar cells and a surrounding layer of flattened elongated cells (Fig. 2). The larger cells contained a large, oval nucleus with a prominent nucleolus and a small amount of pale-staining vacuolated cytoplasm. The external layer was made up of poorly defined elongated cells with large elliptical nuclei. This epithelium rested upon fibrous connective tissue which formed the capsule of the tumor. Numerous septa intervened between the glandular epithelium. Many areas of the glandular tissue were distended into cystic cavities. Here the epithelium was thrown into papillary folds which had a central connective-tissue core (Fig. 2). The lumina of the tubules and cysts were filled with fresh blood and amorphous debris, but the exact contents could not be determined because of the perforation of the tumor during the operative procedure.

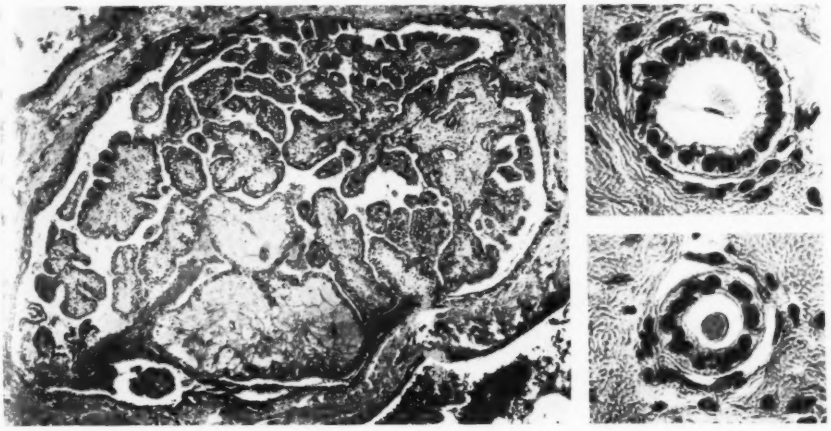

FIG. 2. *Left:* Demonstration of the cystic nature of the tumor and the epithelium thrown into papillary folds. Haemotoxylin and eosin, Zeiss planar 35 mm. *Right:* Cuboidal cells and surrounding flattened, elongated cells constituting the epithelium of the adenoma. ×185.

DISCUSSION

The clinical findings of unilateral deafness and facial paresis combined with roentgenologic evidence of severe erosion of the petrous bone suggested the possibility of a malignant neoplasm at the cerebellopontine angle. However, the chief interest in this case is derived from the unusual tumor that was removed.

From the pathoanatomic point of view, the tumor could easily be confused with a papilloma of the choroid.[3] The cuboidal epithelium regularly arranged on papillae of connective tissue suggested this diagnosis. However, the extradural location made such a diagnosis unlikely. Furthermore, closer examination revealed that the epithelium was not merely composed of a single layer of cuboidal cells as found in choroid. Instead, there was an inner layer of cuboidal to columnar cells and an outer layer of flattened cells analogous to the myoepithelial cells of sweat glands. In some areas the epithelium seemed piled up and appeared to be stratified. The depth of the connective tissue in the capsule and interalveolar septa was much greater than that usually encountered in papillomas of the choroid.

The mechanism by which this tumor arose within the cranial cavity can only be surmised. It has been suggested that the tumor may arise from aberrant glandular

tissues usually found in the external or middle ear.[6] Further investigation of these areas will ultimately have to be done to confirm this hypothesis. Another mode of invasion into the cranial cavity possibly may be malignant degeneration of the tumor and subsequent erosion of the bone. However, there was no definite histologic evidence of malignancy.

SUMMARY

A case is reported of the intracranial occurrence of a ceruminous adenoma which heretofore has been reported as occurring only in the external or middle ear. The tumor was found extradurally in the region of the left internal auditory meatus where it produced a destruction of the petrous portion of the temporal bone, and a paralysis of the 7th and 8th nerves.

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PAPILLOMA OF THE CHOROID PLEXUS

REPORT OF CASE

Everett O. Jeffreys, M.D., and Richard H. Ames, M.D.
Division of Neurological Surgery, Bowman Gray School of Medicine, Winston-Salem, North Carolina

(Received for publication November 17, 1948)

Papilloma of the choroid plexus is a relatively rare intracranial tumor. Less than 100 of these tumors have been reported, and the majority of these were disclosed at autopsy.[2] As far as we have been able to determine from the literature, only 24 papillomas have been diagnosed surgically and of this group less than half the patients survived the immediate postoperative period.

From a pathological standpoint papillomas of the choroid plexus are generally regarded as benign tumors. However, cases are recorded in which histologic evidence of malignancy was present.[3] Secondary tumor implants have been observed as well.[4] In addition, a tendency to recurrence following apparently complete extirpation has been noted.[1,5]

REPORT OF CASE

A 20-year-old white female was admitted to the medical service of the N. C. Baptist Hospital on Nov. 18, 1946 complaining of headache of 6 months' duration. This was mainly frontal and retrobulbar in location and was accompanied by gradually progressive blurring of vision. At times, when headache was particularly severe, there had been nausea and vomiting. She also had noted from time to time diplopia, vertigo, and scotomata.

Examination. The patient was alert, cooperative, and in no acute distress. Positive findings were: bilateral chronic papilledema, weakness of the external rectus muscle on the right, generalized muscular hypotonia with depression of the tendon reflexes, and marked ataxia.

Routine blood and urine studies were within normal limits. Blood serology was negative. Skull films were normal. Visual acuity was 20/40 in each eye. Visual fields showed minimal generalized constriction with gross enlargement of the blind spots bilaterally. Vestibular studies suggested a midline posterior fossa lesion.

The patient was transferred to the neurosurgical service.

Operation. On Nov. 29, 1946, ventriculography was carried out. This disclosed dilatation of the lateral and 3rd ventricles with a complete obstruction at the distal portion of the aqueduct.

A suboccipital craniectomy was performed immediately under general anesthesia. Following the usual operative exposure, which included resection of the posterior arch of the atlas, it was seen that the cerebellar dura was under extreme tension. Ventricular tap relieved the tension to an extent that a dural flap could be reflected without difficulty. The vermis was considerably widened and its folia were obliterated. Projecting from the foramen of Magendie was a tongue of reddish papillary tumor tissue which almost completely filled the cisterna magna. Additional exposure of the tumor mass was attempted by a midline incision into the cerebellar vermis, which proved to be only a few mm. in thickness. Immediately, an irregular cauliflower-like mass, some 4 or 5 cm. in diameter, extruded itself from the 4th ventricle. Considerable hemorrhage occurred at this point both from the surface of the tumor and from the tumor bed. The tumor was lifted from the field without finding a point of attachment. In retrospect, it seems likely that the vascular connections were torn when the tumor mass extruded spontaneously. In any event, as soon as the tumor was removed, bleeding abruptly stopped except for one or two small points in the roof of the ventricle. These were easily coagulated and the field was observed to be dry.

Inspection of the 4th ventricle revealed a tumor nodule, 3 mm. in diameter, firmly implanted in the floor of the ventricle. It was felt that this nodule could not be successfully removed in view of its location and attachment. No other bits of tumor could be seen. Fluid could be seen flowing from the distal end of the aqueduct. The dural flaps were then replaced but not sutured and the wound was closed in the usual manner. The patient was returned to the ward in fairly good condition.

Pathological Report (Dr. W. C. Thomas). "An irregular mass of tissue measuring $4 \times 4 \times 3$ cm. The tissue externally is made up of a grayish-yellow to a grayish-pink type of material, having no definite architectural features. The tissue is found to be easily fragmented and very friable. On attempting to section this tissue, it fragments into numerous pieces, and fragmented portions of this tissue are submitted for sectioning.

"On microscopic examination, this tissue is seen to be made up of many small papillary processes supported on a delicate fibrous stroma containing small capillaries (Fig. 1). The papillary processes are made up of cells which appear to be low columnar or tall cuboidal type cells, and the characteristics of most of the nuclei are similar, although one sees an occasional nucleus which is small and dark. *Diagnosis:* Papilloma of Choroid Plexus."

The *postoperative course* was stormy, with marked fluctuation of temperature and pulse during the first 3 days. Following this, the vital signs became stabilized but she remained quite stuporous. Her stupor persisted, necessitating tube feedings and intravenous fluids, until Jan. 19, 1947, when she roused sufficiently to speak and to take small amounts of nourishment by mouth. Her improvement continued and she was finally discharged from the hospital on Feb. 27, 1947, showing considerable residual cerebellar signs.

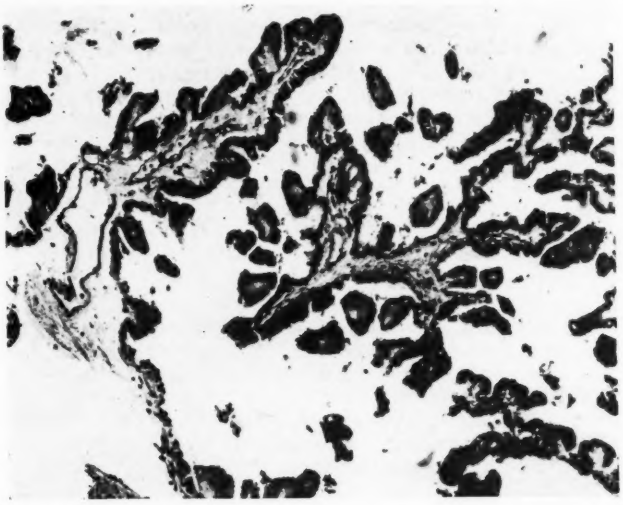

FIG. 1. Photomicrograph of tumor. Hematoxylin and eosin, ×100.

Prior to discharge she was given a total of 4000*r* to the region of the 4th ventricle in the hope that the remaining tumor nodule might be radiosensitive.

During the following 6 months, the patient's nystagmus and ataxia gradually cleared and her visual acuity improved. She has remained in good health except for pneumonitis, 1 year ago, which responded promptly to antibiotics. She was last seen on Nov. 1, 1948, at which time the operative defect was found to be soft and pulsating. Neurologic examination was entirely negative save for minimal secondary optic atrophy bilaterally.

SUMMARY

A case of papilloma of the choroid plexus of the 4th ventricle with secondary implantation in the floor of the ventricle is reported. In spite of incomplete extirpation of the tumor the patient remains well 23 months after operation.

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FULL THICKNESS AUTOGENOUS NERVE GRAFT

REPORT OF A CASE*

I. JOSHUA SPEIGEL, M.D.

Neurosurgical Service, Michael Reese Hospital, and Department of Neurology and Neurological Surgery, University of Illinois College of Medicine, Chicago, Illinois

(Received for publication November 30, 1948)

The bridging of large nerve defects constitutes one of the major problems in peripheral nerve surgery. When the surgeon has exhausted every maneuver to overcome the defect without success, the only alternative is nerve graft, if the nerve is to be saved at all. It is becoming increasingly evident that, in man, autogenous grafts have by far the best chance of survival.[1,2,3,4,5,6,7]

As pointed out by Björkesten,[1] in cases where a full thickness autogenous nerve graft is available, the apposition is much easier technically than in those cases where a cable graft needs to be used. He further points out the fact that full thickness autogenous grafts can be employed only in the presence of a severe lesion of two nerves, simultaneously preventing direct suture of either.

In view of the infrequency of such cases, and the evident need for analysis of all cases so treated, the following case report is presented.

CASE REPORT

A 26-year-old infantry officer was struck by a machine-gun bullet in the middle 3rd of the right thigh on 28 Aug. 1944. Immediately following this, there developed complete paralysis of the tibial and common peroneal components of the right sciatic nerve. About 2 weeks after the injury severe burning pain developed in the dorsum and heel of the foot. This was diagnosed as causalgia, and a lumbar sympathetic block with 1 per cent novocain was attempted. The patient experienced a severe novocain reaction, and the results of the block could not be evaluated. He refused further block, and in view of this, sympathectomy was not advised. Over a period of 4 months the pain gradually disappeared. On 24 Oct. 1944, the sciatic nerve was explored by another surgeon, and an 8 cm. defect between the nerve ends was found. The nerve ends were freshened and approximated with numerous interrupted tantalum wire sutures "under considerable tension." A tantalum cuff was applied. No hip spica was applied, but the knee was put up at 90° in a plaster cast, which was removed in 5 weeks.

Eight months later this patient came to the attention of the writer. He still had complete motor and sensory paralysis of the common peroneal and tibial nerves. There was no advance of the Tinel's sign from the middle of the thigh. Observation of the tantalum sutures in the roentgenograms of the involved thigh revealed an 8 cm. dehiscence of the nerve ends.

Operation. On 29 June 1945, under endotracheal anesthesia, the old mid-thigh incision was reopened and the tantalum cuff surrounded by heavy scar tissue was exposed and removed.

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* Presented before the Chicago Neurological Society on October 8, 1946.

The proximal neuroma and distal glioma were then found. A distance of 8 cm. separated them. Both segments were resected to normal-looking funiculi on both sides, making a total defect of 9.5 cm.

Because the hiatus between the severed nerve endings of the sciatic nerve was too great for primary suture, the common peroneal portion of the sciatic nerve in the distal segment was sacrificed to act as an autogenous nerve graft for the tibial portion. The common peroneal nerve was resected to the head of the fibula, producing a graft 10 cm. in length. The graft was then placed between the tibial nerve ends with the proximal end of the graft directed proximally. Approximation at both ends was without tension using interrupted sutures of fine, tantalum wire on an atraumatic needle. Closure was effected with black silk in layers and a hip spica applied with the hip slightly extended and the knee in flexion.

Course. By gradual degrees the knee was returned to full extension over a period of 6 weeks, beginning the 3rd week. There was no change in the Tinel's sign in the first 3 months postoperatively. The patient was not seen until August, 1946. At this time examination revealed returning function in the soleus and in the two heads of the gastrocnemius. Tinel's sign was present along the course of the tibial nerve in the middle 3rd of the calf, and sensation was still absent in the entire tibial nerve area. Intensive physical therapy was prescribed.

Now (October, 1948) the contractions of the gastrocnemius and soleus are sufficiently powerful to enable the patient to stand on his toes. No function has returned to the flexors of the toes. The sole of the foot (except for the saphenous area) is anesthetic. Sensation is almost normal in the remainder of the foot with the exception, of course, of the common peroneal area which, in its peripheral portion, is hypalgesic and hypesthetic, and in its central portion is anesthetic. The patient wears a strong foot-drop brace and walks with a steady, sure gait.

SUMMARY

A case is reported in which complete return of soleus and gastrocnemius function and moderate sensory return (with the exception of the sole of the foot) were observed, $3\frac{1}{2}$ years following the bridging of a $9\frac{1}{2}$ cm. gap in the tibial nerve by employing the distal segment of the patient's simultaneously injured common peroneal nerve as an autograft.

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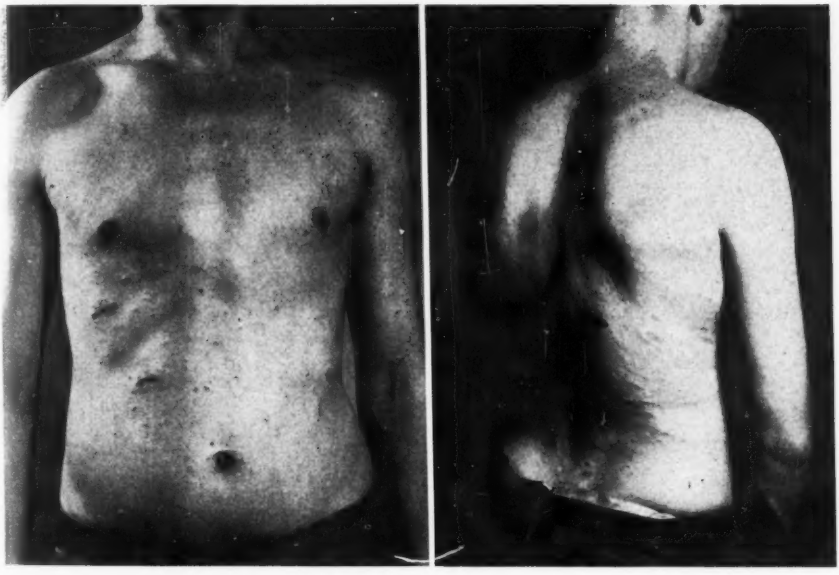
LATERAL INTRATHORACIC MENINGOCELE*

GLEN O. CROSS, M.D., JAMES R. REAVIS, M.D., AND WILLIAM W. SAUNDERS, M.D.
Veterans Administration Hospital, San Francisco, California

(Received for publication December 20, 1948)

Eight previously reported cases of lateral intrathoracic meningocele, only 3 earlier than 1948, define a uniform syndrome. A patient with cutaneous manifestations of von Recklinghausen's disease complains of mild or severe, long-continued back pain. Roentgen examination shows a sharply defined paraspinal tumor usually with local spinal bone destruction. The diagnosis is almost invariably paraspinal or intraspinal neurofibroma until the surgeon finds a cyst communicating freely with the dural sac. Death from sepsis or from shock has followed on 5 such occasions.

Our case is thought to be worthy of presentation because the outstanding finding of spinal destruction was readily proved to be secondary to dural lesions, including multiple meningoceles, by air and pantopaque myelography.



FIGS. 1 and 2. Photographs of patient showing cutaneous manifestations of von Recklinghausen's disease.

CASE REPORT

A 23-year-old white male, a student watchmaker, entered this hospital Mar. 23, 1948 with complaints of pain in the back at the level of the inferior angle of the scapula radiating

* Published with permission of the Chief Medical Director, Department of Medicine and Surgery, Veterans Administration, who assumes no responsibility for the opinions expressed or conclusions drawn by the authors.

around the trunk to the right costal margin. He reported being in good health (without deformity of the back) until December 1944, when in combat in Germany, he was blown from the top of a tank by a land mine explosion and rendered momentarily unconscious. He noticed mild back pain at that time but did not seek medical attention and continued in combat. The pain continued about a week, then subsided to recur several times during the next few months, causing him to seek relief at various medical aid stations. In April 1945 he was wounded in the right leg by shrapnel, and was hospitalized. Again, following his recovery he

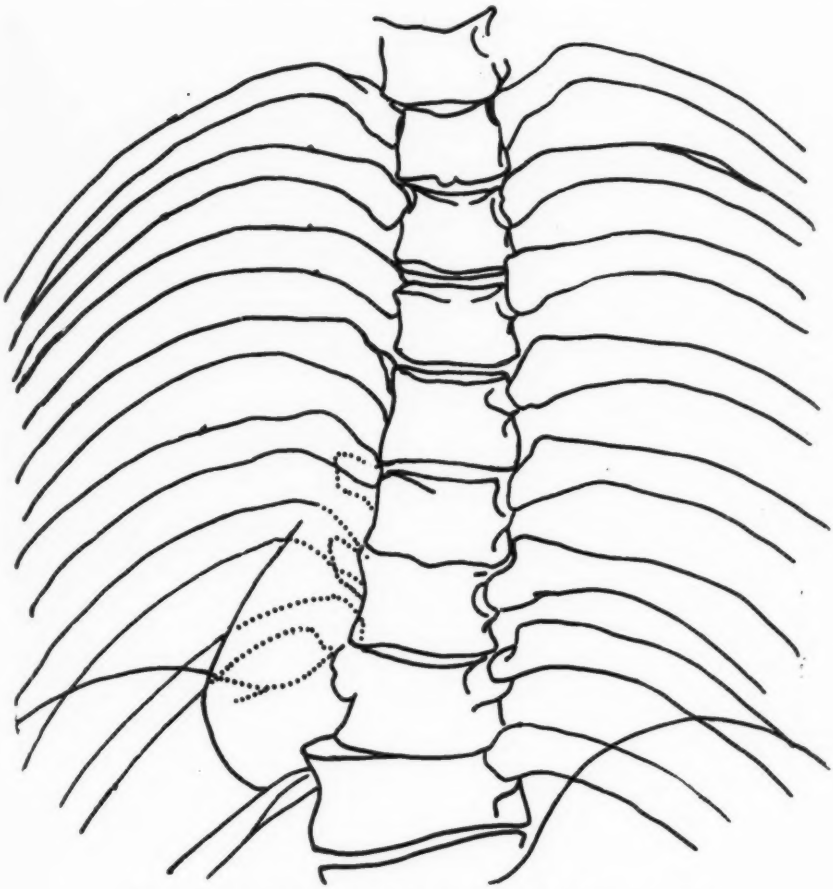


FIG. 3. Illustrating protrusion of the tumor into the right mediastinum.
(Tracing of roentgenogram on p. 424.)

returned to combat. After his discharge from the army on points in January 1946, he worked variously as a bookkeeper and as a laborer in a cannery doing heavy lifting. This latter type of work caused no increased difficulty with his back. He entered watchmaking school in September 1947. In October 1947 he began to have more frequent and more severe pain in the back and right side. He saw Dr. Howard Black of Sacramento, California who, in view of his von Recklinghausen's disease and his symptoms, referred the patient to this hospital to rule out a neurofibroma involving the vertebral column.

Physical Examination. The patient's body was covered with pea-sized soft subcutaneous neurofibromata. There were numerous café au lait spots over the trunk and extremities and a large, black, seed-bed lesion paravertebrally at T9 on the right (Figs. 1 and 2). There was moderate kyphosis of the thoracolumbar spine, most prominent between T11 and L2, which was associated with a right convex scoliosis. The lumbar spine, hips, knees and ankles had a full range of painless motion. Forward bending while sitting caused an increase in the girdle

pain while hyperextension relieved it. Scoliosis was increased on forward bending. It was possible to correct the lateral curvature of the spine but this did not decrease the rotation of the spine towards the right. The left iliac crest was 2 cm. higher than the right.

Neurological Examination. The cranial nerves were entirely normal. The vertebral spinous processes were not tender to pressure or fist percussion. An area of hypalgesia and hypesthesia was found on the right side corresponding to the spinal segments T7 to T11. The tendon reflexes in the lower extremities were mildly hyperactive. No pathological reflexes were elicited.

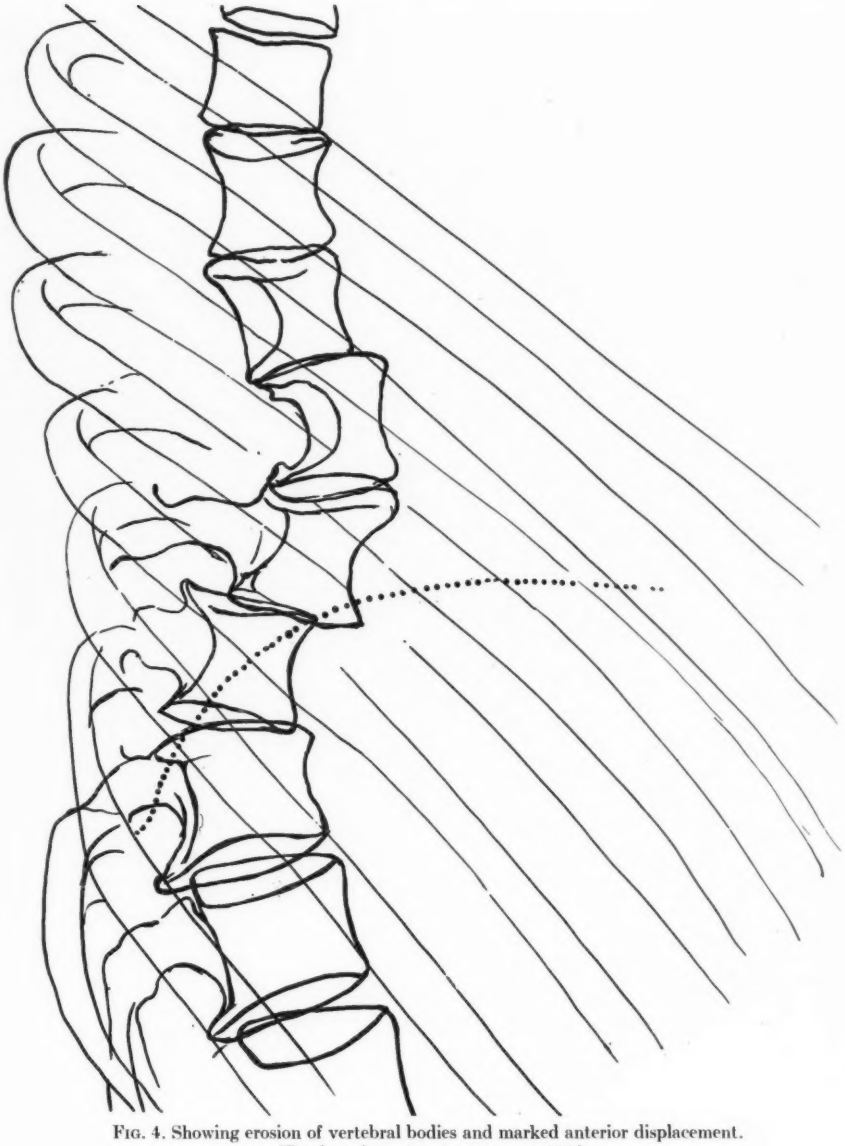


FIG. 4. Showing erosion of vertebral bodies and marked anterior displacement.
(Tracing of roentgenogram on p. 426.)

Position sense and vibratory appreciation were normal. There was no weakness of the anal or vesicle sphincters. There was no demonstrable weakness of the lower extremity musculature. Pulses in the lower extremities were normal.

Spinal puncture revealed a CSF pressure of 140 mm. of water and the manometric studies

were normal. No cells were demonstrated in the spinal fluid; total protein was 23 mg. per cent. Wassermann and Kahn were negative; colloidal gold curve was 1111100000.

Roentgen Examination. Roentgenograms of the spine (Figs. 3 and 4) demonstrated a soft-tissue lesion expanding the thoracic and upper lumbar neural arches and intervertebral foramina. It was most prominent at T10 and T8. Here the pedicles were disrupted, allowing displacement of the bodies of the vertebrae from the neural arches to form a short anterior arc. The dorsa of the bodies were deformed, apparently by pressure erosion. They were smoothly arched between the intervertebral discs, which were much less involved. Subluxation with rotation toward the left had occurred at T10, accompanied by thinning of the intervertebral disc. A sharply defined rounded soft-tissue mass projected into the right lower lung field for a distance of 4 cm. laterally and 8 cm. ventrally from the spine, centering at T10. The pedicles were narrowed or absent in the midthoracic region and the interpeduncular distances were increased from T3 to L2.

Because of these findings and the clinical evidence of von Recklinghausen's disease, the possibility of an intrathoracic meningocele was considered. Myelography was done demonstrating the ready entry of oil into the paravertebral mass at T10 and into masses on either side of the T11 intervertebral disc. The oil reached to within 3 mm. of the periphery of the intrathoracic mass, and close to the dorsa of the displaced, deformed vertebral bodies. Air

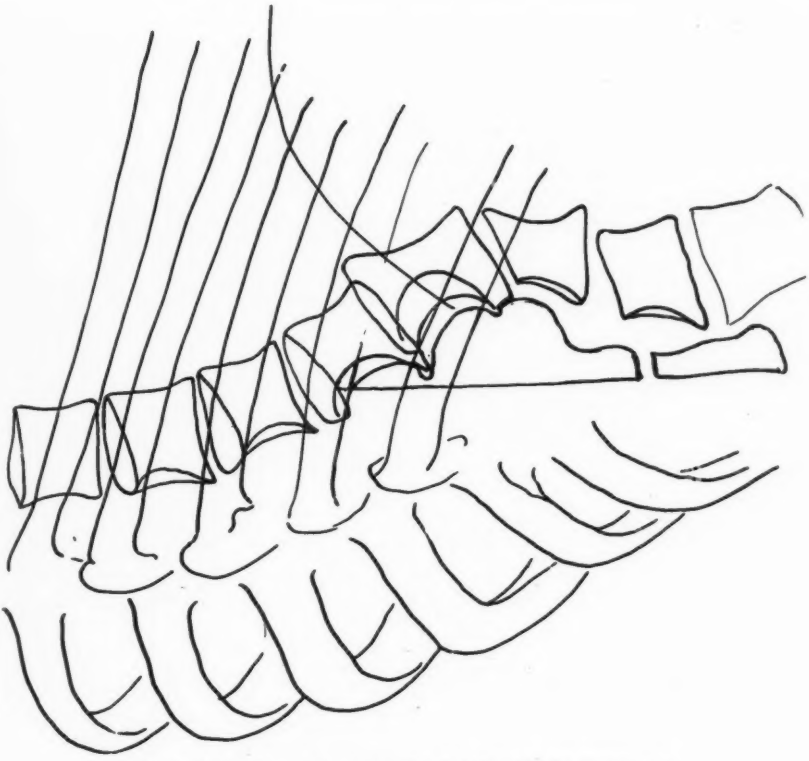


FIG. 5. Air myelogram demonstrating fluid level in the meningocele.
(Tracing of roentgenogram on p. 428.)

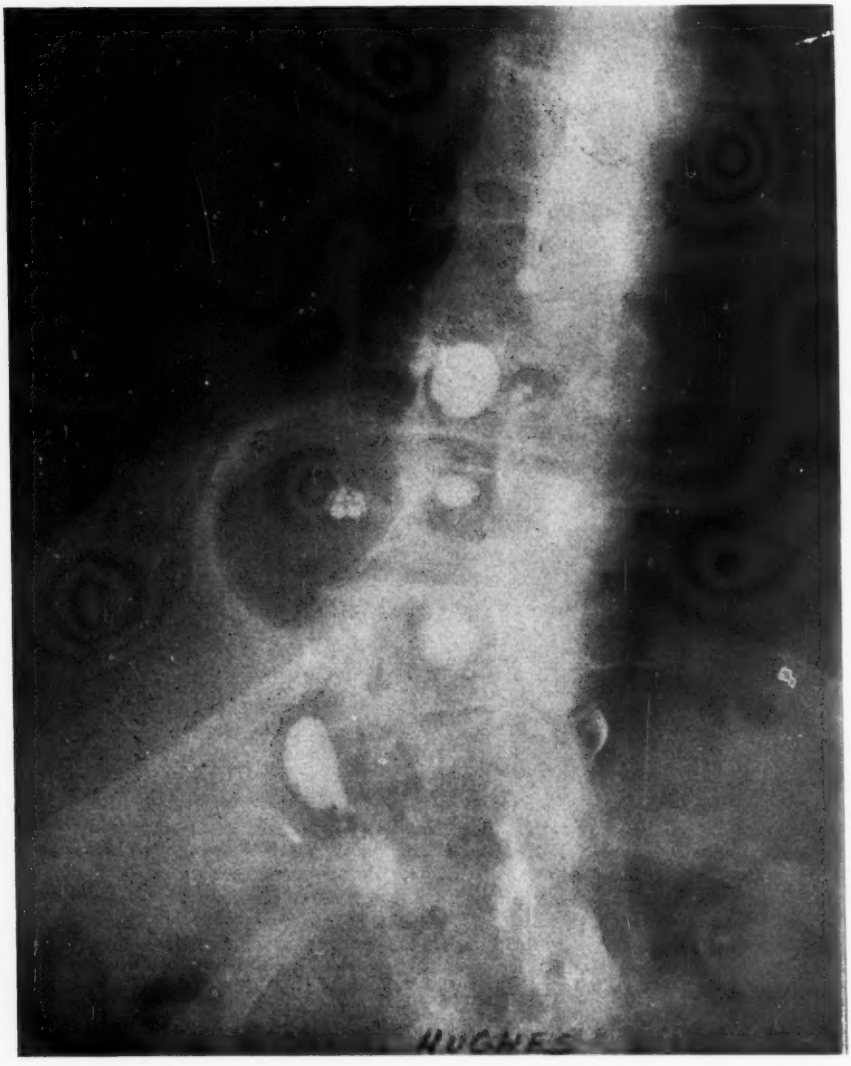
myelogram (Figs. 5 and 6) was then done with the patient lying in a prone position, with pillows placed to elevate the pelvis; 250 cc. of CSF were replaced by an equal volume of air in 10 cc. steps. This revealed the extent of the fluid spaces communicating with the spinal subarachnoid space. Other laboratory studies, including gastrointestinal and urographic examinations, were not contributory.

In view of the high mortality rate attending operative procedures on this type of lesion, it was decided to try conservative treatment. The patient was fitted with a Boldrey brace which relieved his girdle pain. He has been discharged from the hospital and is being followed in the neurosurgical follow-up clinic.

DISCUSSION

As pointed out by Welch *et al.*[6] and by Byron,[2] the usual diagnosis that is made when a patient with von Recklinghausen's disease is found to have a mediastinal tumor, is "neurofibroma." Schüller and Uiberall[4] were the first to make the diagnosis of intrathoracic meningocele preoperatively by the use of myelography. As far as we can ascertain air myelography has not been previously employed in the study of these tumors.

Five of the intrathoracic meningoceles known to us have been associated with



gross von Recklinghausen's disease. Two other case histories make no mention of the condition; however, the *forme fruste* of the disease is not ruled out. This overwhelming correlation created a strong presumption of etiologic relationship, the nature of which is not clear.

Kyphosis in patients with von Recklinghausen's disease has frequently been noted. Few of these patients have spinal cord tumors. Most if not all of the patients with meningoceles had kyphoscoliosis of some degree. One patient[1] had a spinal

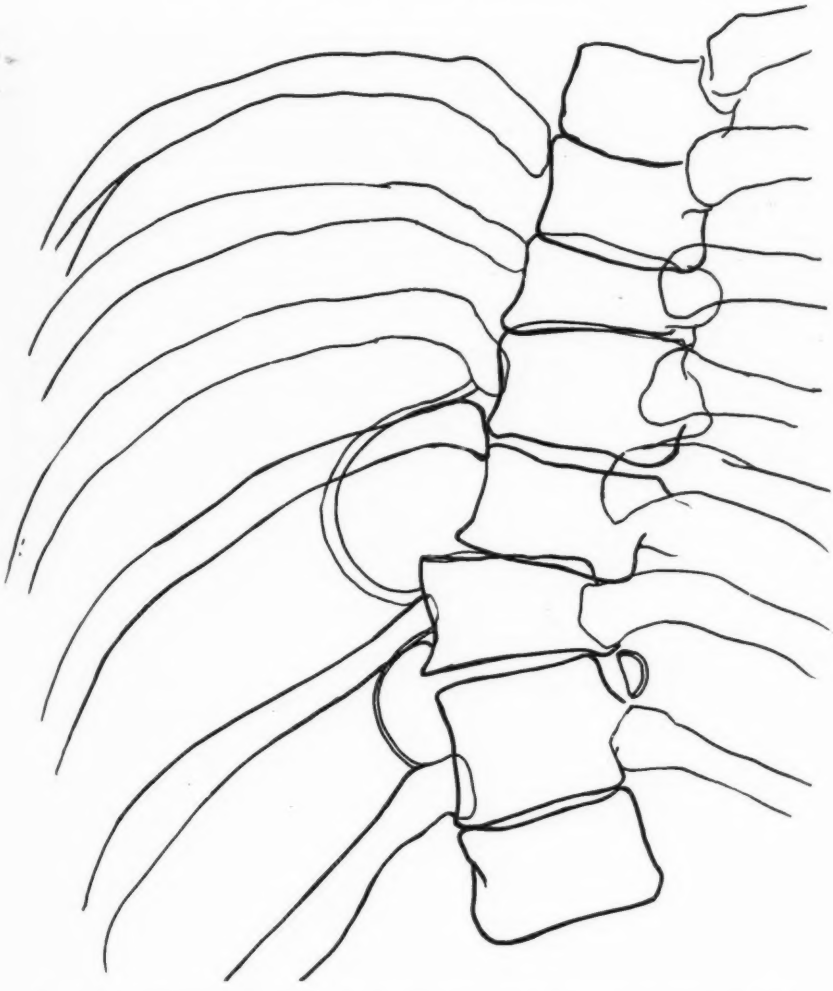


FIG. 6. Air myelogram demonstrating the meningoceles. (Tracing of roentgenogram on p. 430.)

fusion for correction of the deformity 11 years before the discovery of the meningocele.

Previous authors speculating on the origin of these lesions have agreed that a developmental defect either of the bone or of the dura precedes the appearance of the meningocele. That the actual herniation may progress is shown by the fact that 2 of the patients were demonstrably without tumor 11 and 5 years previous to the final diagnosis.[1,2,3,4,5]

The bony defects demonstrated in the meningocele cases appear to be the result

of pressure necrosis caused by an expanding lesion, ranging from minimal enlargement of an intervertebral foramen in the case of Sengpiel[5] to elongation and fracture of several neural arches, marked arciform erosion of many thoracic centra and multiple herniations of the dura, as found in our case. No adequate evidence is available to prove whether these deformities preceded or followed the herniations. Examinations reported prove the absence of meningocele but do not prove the absence of bone defects. Even in our patient one can postulate that a marked spinal deformity had existed since birth. Final decompensation with destruction of an intervertebral disc, increased kyphoscoliosis and development of radicular pain followed an adequate injury by a period consistent with the course of traumatic arthritis. Meningocele formation may have been incidental.

Sengpiel *et al.*[5] have suggested that an unusually extended attachment of the dura onto the nerve root sheaths might create a spinal-fluid-filled pouch capable of expanding under increased spinal fluid pressure as would occur in coughing. From our observations we might add that a neurofibroma might distort the dura at the intervertebral foramen with the same results. These mechanisms do not account for the bony deformities, especially in our patient whose lesion is predominantly intraspinal.

Solid and pulsating vascular intraspinal tumors are well recognized causes of such bone erosion. We find it hard to conceive how spinal fluid pressure acting on normal, or even thinned dura could produce the same necrosis. As an alternative to this concept, we suggest that a defect in closure of the dura at an early age might cause hypoplasia of the surrounding bone, which alteration might persist through the growth period to finally result in meningocele formation.

SUMMARY

1. Five of 8 cases of lateral intrathoracic meningocele have been associated with gross von Recklinghausen's disease. This establishes a strong presumption of common etiology.

2. We suggest that these lesions result from early developmental defects of the dura rather than of the bone.

3. Myelography will establish a pre-operative diagnosis and should be done when neurological symptoms are insignificant in proportion to structural changes.

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OLIGODENDROGLIOMA OF THE SPINAL CORD*

JOHN R. RUSSELL, M.D., AND PAUL C. BUCY, M.D.

The Chicago Memorial Hospital and the Department of Neurology and Neurological Surgery, The Illinois Neuropsychiatric Institute, University of Illinois College of Medicine, Chicago

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Oligodendrogliomas comprise 3.5 per cent of the gliomas of the brain, according to the analysis of 254 histologically verified gliomas classified by Bailey and Cushing.[3] In their discussion of oligodendroglioma of the brain, Bailey and Bucy[2] pointed out that this variety of glioma should also occur in the spinal cord, since the oligodendroglia are found in the spinal cord as well as in the brain.

Detailed histological analyses have been made of only a few large series of intramedullary spinal cord tumors. Kernohan, Woltman and Adson[6] in 1931 classified 31 cases of intramedullary spinal cord tumor operated upon at the Mayo Clinic and found 2 (4 per cent) oligodendrogliomas. Up to Jan. 1, 1939, the Mayo Clinic series included 64 intramedullary tumors, 3 of which were oligodendrogliomas, an incidence of 5 per cent.[9] The same group[7] reported 25 gliomas of the conus medullaris and filum terminale, of which 1 was an oligodendroglioma. In 1944 Woods and Pimenta[11] made a study of 30 histologically verified spinal cord gliomas, and classified 1 as an oligodendroglioma. Oljenick[8] has also reported 1 case, giving a total of 6 cases of oligodendroglioma of the spinal cord and filum terminale recorded in the literature.

A careful search of the literature from 1930 to 1947 disclosed an additional 98 intraspinal gliomas, histological classification of which was attempted. The exact pathologic diagnosis is subject to question in some cases, but there was no instance of an oligodendroglioma. Thus oligodendroglioma of the spinal cord is a very rare lesion. For this reason we feel it worth while to report a case in which operation was performed on May 13, 1948. The symptoms were of long duration, and there was an associated scoliosis. This case has been briefly reported by Bucy and Heimburger.[4]

CASE REPORT

History. A. LeC., a white male railway clerk, aged 31, was referred by Dr. Harry Hodges and admitted to the Chicago Memorial Hospital on May 11, 1948.

In 1932, at the age of 15 years, the patient was in an automobile accident, but sustained no injury that was apparent at the time. One month later he noticed protrusion of one of the lower left ribs, and was told that it was fractured. Two years after the accident "twitching movements" occurred in the muscles of the back, and his family noticed that his spine was crooked. At about the same time he began to have periods of clumsiness and numbness in both hands, lasting only a few minutes. Following 5 months of naprapathic treatments in 1937, the patient noted no fibrillations in the back muscles, until they returned in 1940. The "twitches" occurred at a rate of about 40 per minute, were most pronounced when the patient was sitting, and caused his entire body to jerk.

During the next 2 years there developed slowly progressive weakness and awkwardness of the legs and hands. He fell twice, due to weakness of the legs. His gait was peculiar and he carried one shoulder lower than the other.

Examination by an orthopedic surgeon in 1942 revealed a lower thoracic scoliosis with

* Presented before the Chicago Neurological Society, January 11, 1949.

convexity to the right. The left shoulder was higher than the right. Rhythmical contractions of the left sacrospinalis musculature were palpable. A questionable decrease in pain sensation in the lower extremities was noted, but a detailed neurological examination was not recorded. X-ray films of the spine made at that time showed the scoliosis (Fig. 1).

On May 20, 1942, the orthopedic surgeon performed a spinal fusion with a 6 inch tibial bone graft placed on the denuded laminae from the 10th thoracic to the 3rd lumbar vertebra. On June 9, 1942, a 2nd operation was done to correct the malposition of the tibial graft. Following this the wound became infected and drained for 4 months. After recovery from the operation the patient was completely relieved of the twitching in the back muscles. He had no weakness or clumsiness of the extremities, except that he considered the right leg a little weak, and favored it because the tibial bone graft had been removed from that side. In due time, he was able to return to work.

FIG. 1. Composite roentgenograms of spine showing scoliosis in 1942.

In December, 1946, the patient sustained burns on the thighs from a hot radiator, but felt no pain. Following this he had several minor traumata to the legs, without any sensation of pain. In the Spring of 1947, he noticed clonus of the right ankle while playing the organ. After that he had a progressive weakness of the legs, the right being most affected. By the Summer of 1947 he was unable to climb stairs or run, and had great difficulty in walking. In the year before his admission to the hospital, the patient had fecal and urinary urgency, and was incontinent if unable to relieve his urgency at once. He also complained of aching pain in the lower abdomen and back.

Examination. When admitted to the hospital on May 11, 1948, the patient could barely walk with the aid of two canes. His gait was spastic, with a broad base. The Romberg test was positive. He was unable to hop on either foot. There was generalized weakness of both lower extremities, most marked in the hamstring muscles, and greater on the right than on the left. The thighs and calves were markedly atrophic. The tonus of the lower extremities was increased, especially on the right. The knee jerks were hypoactive, but the ankle jerks were hyperactive. Ankle clonus and Babinski's sign were present bilaterally. Spontaneous flexor spasms were noted. The abdominal reflexes were present in the upper quadrants but absent in the lower quadrants. He could not feel pin prick below the 11th thoracic dermatome. Perception of warmth was absent below the 2nd lumbar dermatome, and of cold was present but diminished below the same level. Position sense was diminished in the right foot, and the ability to recognize numbers written on the skin was absent in both legs. Tactile sensibility was normal. No neurological defect was found in the upper extremities or in the cranial nerves. General physical examination revealed a lower thoracic scoliosis with convexity to the right, and a surgical scar from the 9th thoracic to the 3rd lumbar spinous process.

On lumbar puncture the initial CSF pressure was 160 mm. fluid. Bilateral jugular compression gave a rapid rise in pressure to 300 mm., but on release of the compression the pressure fell slowly and hesitantly back to the original level. After removal of 7 cc. of fluid, the pressure

fell to 0 mm. The impression was that a partial CSF block was present. The CSF was clear and colorless, contained 2 lymphocytes/c.mm., and 78 mg. per cent of total protein. The Wassermann reaction was negative and the Lange colloidal gold curve was 0000000000. Examination of the blood and urine revealed no abnormality.

Roentgenograms of the spine showed a scoliosis extending from the 5th thoracic vertebra to the 3rd lumbar. A dense spinal fusion in the thoraco-lumbar region made detailed examination of the spine and of the spinal canal impossible. A chest x-ray showed no abnormality except old fibrotic scarring in the right infraclavicular area.

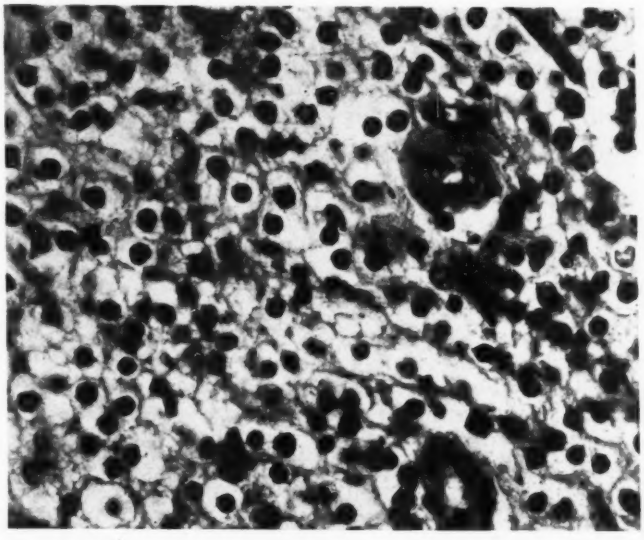

FIG. 2. Photomicrograph of section of tumor stained with hematoxylin and eosin.

Operation. On May 13, 1948 a laminectomy of the 8th, 9th and 10th thoracic vertebrae was made through the old spinal fusion. At the level of the 9th thoracic vertebra there was no epidural fat, and the dura mater was thin and distended. Incision of the dura mater revealed a greatly enlarged yellowish spinal cord. All the usual landmarks were obliterated and the surface vessels were flattened. No fluid could be aspirated from the cord. The left posterior column of the cord was then incised longitudinally for a distance of 3.5 cm. This resulted in exposure of a very gelatinous, pinkish-gray tumor, which herniated through the opening. There was no capsule, and the tumor would not shell away from the cord substance. The upper and lower limits of the tumor were not exposed. A considerable portion, but by no means all, of the tumor was removed with a pituitary rongeur and with suction. The dura mater was left open, and the other soft tissues were closed in layers as usual.

Pathology. Sections of the tumor stained with hematoxylin and eosin show it to consist largely of round or polyhedral cells with large vesicular nuclei, and surrounding clear cytoplasm, giving a box-like appearance to the cells. For the most part these cells are closely packed with no intervening stroma. No mitotic figures or other evidences of malignancy were seen. The walls of many of the blood-vessels are thickened and some are practically occluded by this intravascular proliferation. No calcification was seen. The microscopic appearance of the tissue (Fig. 2) is typical of an oligodendroglioma.

Postoperative Course. The patient was completely unable to move the lower extremities. The sensory level remained at the 11th thoracic dermatome until 3 weeks postoperative, when it had dropped to the 1st lumbar dermatome. On the 9th postoperative day, he began to have a spiking fever, and by the 12th day a spinal fluid fistula was apparent at the lower end of the incision. Lumbar puncture yielded CSF containing 3000 polymorphonuclear leukocytes /c.mm. On the 13th day, a secondary closure of the fistula was done, and no further drainage has occurred. The signs of meningitis subsided with streptomycin, penicillin and sulfadiazine therapy. It is thought that this meningitis was the result of the lighting-up of the old infection which had been present in 1942 and which had been lying latent in the bone graft.

On June 8, 1948, the patient was transferred to the Illinois Neuropsychiatric Institute for rehabilitation from the standpoint of the paraplegia. He has remained paraplegic, but has learned to walk with the aid of braces and crutches. On June 14, 1948 a lumbar puncture revealed an initial pressure of 90 mm. CSF. There was a rapid rise in CSF pressure on jugular compression, with a rapid fall in pressure on release of the compression. The CSF contained 22 lymphocytes/c.mm. The patient had a paradoxical urinary incontinence, with a residual of 500 cc. To correct this, a transurethral bladder neck resection was done on Oct. 2, 1948, following which he developed an automatic bladder, with no residual. He was discharged from the Illinois Neuropsychiatric Institute on Oct 26, 1948.

DISCUSSION

The association of this intramedullary spinal cord tumor with scoliosis has been discussed in another publication.[4] An intramedullary spinal cord tumor that does not destroy the vertebrae probably can cause scoliosis only when it develops slowly in a growing individual. Scoliosis was first observed in this patient at the age of 17. The scoliosis is probably produced by asymmetrical spasm or weakness of the perivertebral musculature innervated by the involved portion of the spinal cord. It may be noted that this patient observed twitching of the muscles of the back at the onset of the scoliosis. Vertebral erosion by pressure of an intraspinal tumor can lead to scoliosis, but this is very unlikely in the case of intramedullary lesions.

Allen and Kahn[1] reported a case of scoliosis produced by an upper thoracic and cervical intramedullary astrocytoma. Their patient, who was 7 years old at the onset of symptoms, had an 8-year history of scoliosis and back pain, with weakness of the left leg for 10 months.

Our patient has an unusually long history of signs and symptoms, dating back 14 years prior to operation. In Kernohan's[9] cases, the duration of symptoms was 8 years, 4 years, and 6 months (personal communication). In the case reported by Oljenick[8] symptoms were present for 2 years. The patient of Woods and Pimenta[11] had symptoms for only 4 months, and died $4\frac{1}{2}$ months after operation, but the tumor tissue showed "relatively malignant cellular characteristics."

Ependymoma is by far the most common intramedullary spinal cord tumor. In the largest recorded series, ependymomas comprise 40 to 50 per cent of the gliomas. All other types of glioma are found in the spinal cord, but the incidence of any one type, such as oligodendroglioma, is low.

Oligodendrogliomas occurring in the brain frequently contain calcareous deposits visible by x-ray examination. No such calcification was present in our case, or in any of the 6 other recorded cases. Weaver[10] reported a lower cervical glioma "moderately cellular and with considerable calcification," but did not specify the cell-type. Gray[5] reported a case in which the roentgenogram showed a spotty calcification within the spinal canal. At operation an intramedullary tumor was found, but no biopsy was taken. Jefferson[5] thought that this lesion was a hemangioblastoma or possibly a very vascular oligodendroglioma.

Kernohan, Woltman and Adson[7] pointed out that in their large series of intramedullary tumors of the spinal cord, conus medullaris and filum terminale, only 3 of the tumors invaded the subarachnoid space, and 2 of these were oligodendrogliomas. No invasion of the subarachnoid space had occurred in our case.

SUMMARY

1. A case of oligodendroglioma of the spinal cord occurring in a 31-year-old white male is presented. There were symptoms of scoliosis for 14 years, followed by progressive motor and sensory loss in the lower extremities.

2. The scoliosis in this case is thought to be due to the intramedullary spinal cord tumor.

3. A review of the literature is given; 6 other cases of primary intraspinal oligodendroglioma have been reported.

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BOOK REVIEW

OBSERVATIONS ON THE PATHOLOGY OF HYDROCEPHALUS. By Dorothy S. Russell. *London: His Majesty's Stationery Office*, 1949, vi, 138 pp., 90 figs. Paper covers. Price, 6 shillings.

Professor Russell's superb monograph takes as its point of view that "the common background of hydrocephalus is formed by the single factor of obstruction, and the immense variety of pathological lesions productive of hydrocephalus have this single feature in common: all create an obstruction at some point in the pathway of the cerebrospinal fluid." In each instance of a large series of cases, Russell has been able to identify the cause of the obstruction, a record hardly approached in most neuropathological laboratories.

The results are discussed on an etiological basis. Of the malformations, particular emphasis is given to stenosis and forking of the aqueduct and the Arnold-Chiari malformation. Abundant evidence is presented that hydrocephalus associated with meningocele is not due to traction resulting from fixation of the spinal cord at the site of the meningocele. A chapter is devoted to a form of progressive gliosis of the aqueduct leading to gradually increasing hydrocephalus. The condition is of uncertain etiology, though Russell presents some evidence that it may be the result of inflammation. The frankly inflammatory lesions are treated thoroughly. Russell has gathered evidence that certain instances of hydrocephalus are due to clinically unrecognized meningeal inflammation a long time previously. Dural sinus thrombosis with survival is regarded as a cause of mild or moderate hydrocephalus, perhaps reversible in part. Neoplasms as a cause of hydrocephalus are discussed briefly but adequately. There is an especially valuable chapter on the anatomical effects of hydrocephalus *per se*.

The distinction of this monograph depends in part on the meticulous study of a large material; on fair and dispassionate presentation of all points of view; on the breadth of understanding which unites neuropathological investigation with relevant clinical and experimental data; and on the distinguished literary style. The illustrations are very well chosen and clearly reproduced. The paper and printing are satisfactory, but so valuable a monograph would seem to merit a more secure binding.

This book is by far the best available discussion of hydrocephalus. It is indispensable to anyone concerned with this condition, whether neuropathologist, neurosurgeon, pediatrician or roentgenologist. The reviewer would like to point out also that those interested in the best methods for presenting scientific material could have no better model than this monograph.

ORVILLE T. BAILEY, M.D.

METASTASIZING CEREBELLAR TUMORS

THE DIFFICULTY IN DISTINGUISHING BETWEEN MEDULLOBLASTOMA AND NEUROBLASTOMA

ROBERT P. BARDEN, M.D., AND FREDERIC H. LEWEY, M.D.

Department of Radiology and the Pathological Laboratory of the Neurosurgical Service, Hospital of the University of Pennsylvania, Philadelphia, Pennsylvania

(Received for publication March 19, 1949)

IT is now 20 years since one of the first reports in the literature of cerebellar neoplasm with tumors in distant bones was presented from this clinic.[10] In the ensuing years, evaluation of many of the clinical and radiologic problems attending cerebellar medulloblastoma has been pursued based on experiences with patients passing through our neurosurgical clinic.[5,9] As a result, additional information has become available which warrants re-examination of the concept that medulloblastoma may occasionally metastasize to extraneural structures.

Although it is generally agreed that these tumors remain localized to the central nervous system, sporadic case reports of skeletal and visceral dissemination, in addition to the one mentioned above, have appeared in the literature.[8,13,16] When all of these are critically appraised, it appears that the group of the "medulloblastoma" is not so unequivocally defined as it was originally thought to be. The term "medulloblast" was coined by Bailey and Cushing[1] to designate a hypothetical immature, bipotential cell which may develop into neuroblasts or glioblasts. This concept implied the possibility that in some of these tumors, neuroblasts prevailed, in others, glial elements. A similar thought was expressed in the opinion of two opposing schools of thought as to the origin of the type cell of these tumors. Penfield[11] and his co-workers suggest that a group of embryonal cells commonly found in the anterior medullary velum give rise to the formation of the medulloblastoma, whereas Stevenson and Echlin,[15] and others derive them from the marginal layer of embryonal granular cells in the cerebellar cortex which gradually disappear in the third year of life. These authors prefer the name granulocytoma to medulloblastoma. Be this as it may, the consensus of opinion today is that there exists a common tumor of childhood of a particular type, located primarily in the cerebellar vermis, though later extending into the hemisphere. This tumor is extremely radio-sensitive in the beginning, without being completely destroyed by any present method of radiation. With repeated irradiation, it becomes increasingly insensitive, and eventually, lethal, after a course of from 1½ to 3 years. There seems to be no doubt that the tumor represents clinically, pathologically, and radiologically a well defined disease entity independent of the name given to it.

The ambiguous nature of the characteristic cell of this tumor, on the other hand, raises the question as to whether some of these neoplasms may not be part of the group of so-called sympathicoblastomas. This means neuro-

blastomas of the sympathetic nervous system, and would open the possibility that a primary tumor in the adrenals, the sympathetic chain, or anywhere else in the body in a sympathetic ganglion could metastasize into the brain and the bones. On the other hand such tumors might originate in various organs independently of one another or take their origin from sympathetic or other immature nerve cells in the brain and metastasize from there, although no precedent is known for this sequence of events.

It seems, therefore, important to review the subject of disseminating cerebellar tumors, using as a basis for discussion 1 of our early cases and 2 others recently observed.

CASE REPORTS

**Case 1.* A white female, aged 32, was admitted to the service of Dr. Frazier at the University Hospital on Nov. 1, 1919, her chief complaint being staggering gait. The symptoms and signs, which indicated cerebellar dysfunction and increased intracranial pressure, began about July of that year.

A 1st stage suboccipital craniectomy was performed several days after admission. Subdural tension was extreme, but on account of the poor condition of the patient, the 2nd stage was delayed for 2 weeks. At that time, the tension had subsided somewhat, and the only abnormality encountered was a somewhat enlarged right cerebellar hemisphere. Fifty mg. of radium were inserted into the cerebellar hemisphere and left there for 18 hours. Two weeks later, the patient was discharged in fair condition. Pathologic study of a small amount of tissue removed proved it to be edematous cerebellar tissue.

In January, 1921, another operation was necessary, and this revealed a large, soft, friable, grayish-red tumor covering the left cerebellar hemisphere. As much of this as possible was removed, and 50 mg. of radium were applied for 11 hours. Subsequently, the patient received 5 radium applications. The pathologic diagnosis at that time was "glioma (medulloblastoma)." Five months after discharge, she returned for more radium applications. She did not seem so well as previous to the operation. However, she had neither headache nor pain. For approximately ½ year, the condition remained about the same, and then there developed pain over the sacrum and shooting pains in the legs.

She did not return to the hospital, however, until October 1923, at which time there was severe pain in the back and shoulders, persistent edema of both legs, and emaciation. Whereas physical examination had previously given negative results except for the neurologic observations, the patient now appeared cachectic. There was a visible and palpable mass just above the left buttock over the sacro-iliac joint, and the back of the lumbosacral region was discolored. Rectal examination revealed extreme tenderness in both sacro-iliac joints. Roentgenograms of the pelvis showed evidence of an extensive growth involving both iliac bones, the right side of the sacrum, and probably the upper part of the right femur (Fig. 1). Roentgenograms of the lower part of the thoracic region and chest disclosed a large shadow to the right and in front of the spine and behind the heart which might have been a tumor originating in the sympathetic chain. Evidence of spinal involvement in this region was not present.

A cordotomy was performed at the 5th thoracic level to control her pain. Upon

* Previously reported by Pendergrass and Wilbur.[10]

discharge from the hospital there was little pain in the legs, although some pain was still present in the region of both hips. The patient died at home and postmortem examination could not be obtained.

Comment. The original histologic diagnosis of the tumor removed at operation in 1919 was "glioma (medulloblastoma)." These sections were reviewed in 1942 and the diagnosis was changed to neuroblastoma (Fig. 2). If this diagnosis were accepted it would remove 1 of the cases of so-called metastasizing medulloblastoma from the literature.

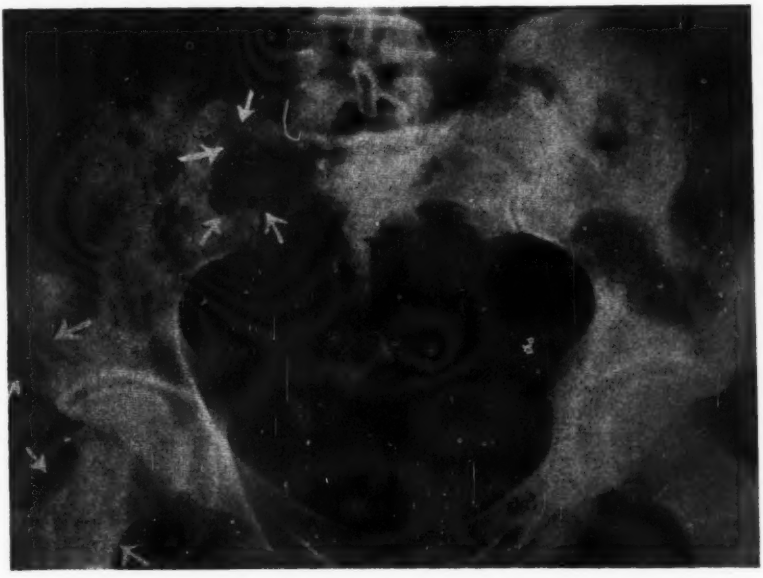

FIG. 1. Case 1. (From *Arch. Neurol. Psychiat., Chicago*, 1928, *19:* p. 443.) "A metastatic growth involving both wings of the ilium and both femora, especially the right, and the right wing of the sacrum. Unfortunately, a large amount of gas tends to confuse the true areas of metastases, which can be easily differentiated in the original roentgenogram."

Case 2. A white male, aged 13 years, was admitted to the University Hospital on Dec. 28, 1938 with a history of severe headache for 3 weeks and intermittent vomiting for 2 weeks. There was also some blurring of vision and difficulty in walking. Examination showed left-sided incoordination, nystagmus, and evidence of increased intracranial pressure. Operation resulted in the removal of a large tumor from the left cerebellar hemisphere.

The tumor was originally called "medulloblastoma" with reservations. On recent detailed histological examination it was noted that the tumor cells were arranged in parallel rows separated from one another by reticulin fibers. Both cells and fibers coiled up in the shape of large whorls in some places and formed pseudorosettes in others. The reticulin fibers sprang from the blood vessels. The majority of the cells were of the larger variety. Their nucleus was irregularly shaped, bright,

its chromatin often assembled at opposite poles. Cytoplasm was scant. Most of the cells were apolar; some had one, occasionally long but usually short, prolongation. No mitoses were found (Fig. 3).

The patient received roentgen therapy during February and March 1939 with a total of 2000 r into the mid-portion of the brain. A 2nd course of therapy was given in July and August, 1939 with a total tumor dose of 2000 r. He remained well until November, 1941, when he began to complain of severe pain in the arms and legs and presented himself with a swelling of the left thigh and an enlargement in

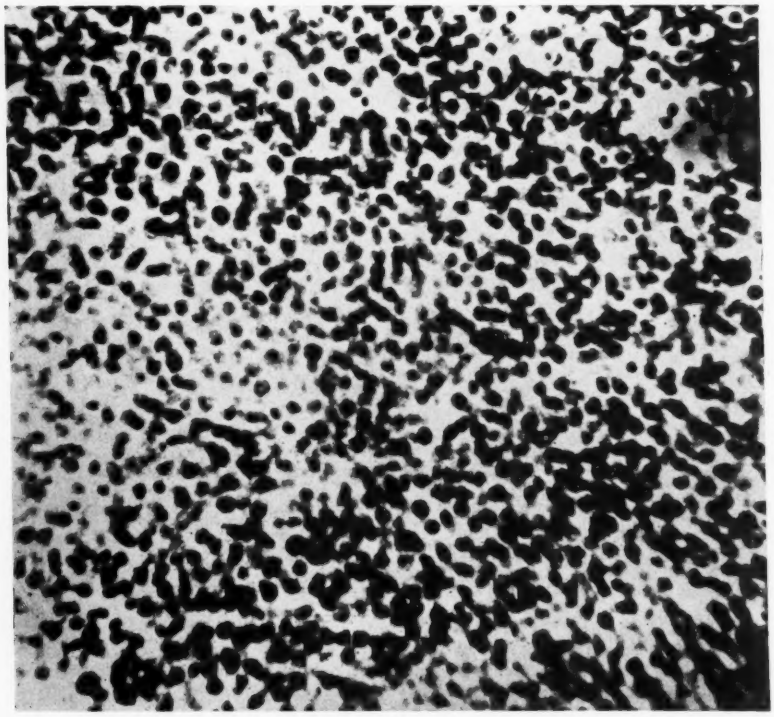

FIG. 2. Case 1. (From *Arch. Neurol. Psychiat., Chicago*, 1928, *19:* p. 442.) "Photomicrograph of tumor which was diagnosed glioma (medullary blastoma)."

the left groin. Although the initial explanation for the pain in the extremities was that the patient had seeding of the recurrent tumor along the spinal cord, roentgenograms of the left femur showed a large destructive lesion in the bone (Fig. 4). The diagnosis was then changed to osteomyelitis. An incision was made in the femur and pus, under pressure, was released.

The histologic diagnosis on the biopsy material was a malignant bone tumor and the consensus of opinion of several pathologists was that it might be classified as a Ewing's tumor. The bone was found to be invaded by strands of tumor cells. The

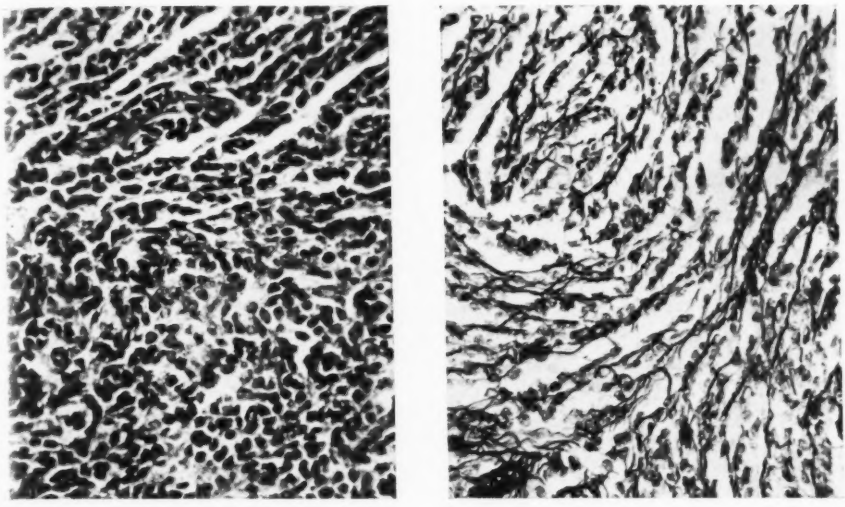

FIG. 3. Case 2. Cerebellar tumor (medulloblastoma or neuroblastoma). *Left*, Hematoxylin and eosin, ×300; *right*, Laidlaw. The cells are arranged in rows which are separated by reticulin fibers springing from the blood vessels. Both cell rows and reticulin fibers coil up to form large whorls which may simulate pseudorosettes. The cells of this tumor are more mature and better differentiated than those of Case 3. (Compare with Fig. 6.)

FIG. 4. Case 2. Roentgenograms of the left femur of a 13-year-old boy previously treated for cerebellar medulloblastoma. Irregular destruction of cortex and medullary cavity, periosteal new bone formation and a large soft tissue mass, all suggest a malignant bone tumor. The original diagnosis from biopsy specimen was Ewing's tumor.

type cell contained an irregularly shaped, elongated nucleus surrounded by a small amount of cytoplasm which tapered out on the two opposite ends of the nucleus. The prolongations formed a loose network. Occasionally, a few cells were arranged in rosette shape (Fig. 5).

Pain was temporarily relieved by further roentgen therapy, but the patient died in September, 1942. Postmortem examination was not obtained.

Comment. This patient exhibited the classical clinical picture and gross and microscopic pathology of medulloblastoma (Fig. 3). His postoperative

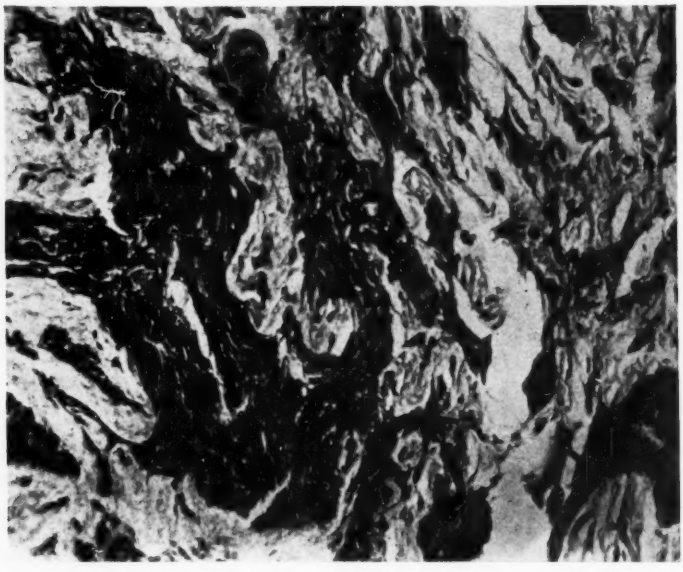

FIG. 5. Case 2. Bone tumor (Ewing's tumor or neuroblastoma). Trichrome, ×440. Invasion of bone by strands of irregularly shaped cells with elongated nuclei and tapering bipolar prolongations. At the upper margin suggestion of rosette formation.

course and response to radiation therapy was exactly as expected until the late complication occurred. It was unfortunate that no autopsy was obtained, but from the evidence of the biopsy of the femur (Fig. 5), it must be assumed that the patient either had two separate tumors of extremely similar histologic pattern, or that the medulloblastoma metastasized, or that both the cerebellar and the bone tumors were part of a metastasizing neuroblastoma (vide discussion).

Case 3. A white male of 14 years was admitted to the University Hospital in May, 1945 because of severe headache, dizziness and loss of equilibrium. In June, 1945, a small midline cerebellar tumor was removed and the pathologic diagnosis was a highly differentiated medulloblastoma.

The tumor showed a definite pattern. The background was formed by a loose

arrangement of small, round, almost bare nuclei with a few short prolongations. Some of the nuclei were dark and round like lymphocytes, others larger, more irregular in outline, with only a few chromatin dots. Many nuclei were carrot-shaped. The amount of cytoplasm increased with the size of the nucleus and surrounded the nucleus without prolongations or ended in one, occasionally bifurcating, prolongation. This diffuse background was permeated by strands of cells in fish-school

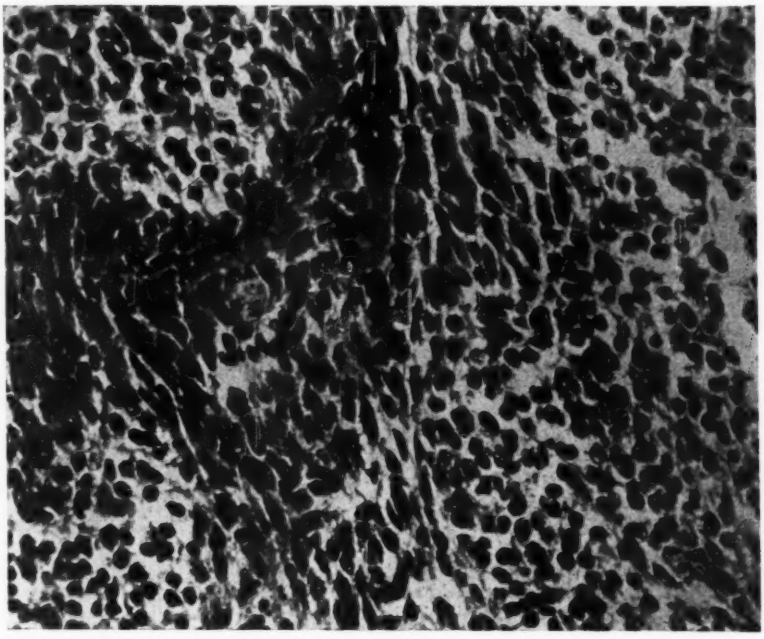

FIG. 6. Case 3. Cerebellar tumor (medulloblastoma or neuroblastoma). Hematoxylin and eosin, ×470. Strands of medium-sized, oblong cells stream through a diffuse accumulation of smaller, often lymphocyte-like almost bare nuclei, loosely arranged. No reticulin or collagen is present. (Compare with Fig. 8.)

arrangement. These cells were considerably larger, their nuclei multiform and bright though with a tendency to elongation, often on one end only. They were tighter packed than the background cells, sometimes in rows but not interconnecting with one another. No reticulin or collagen had been laid down (Fig. 6).

During August, 1945, roentgen therapy was administered to the head with a total of 5,000 r delivered to the midportion of the brain. Another course was given in March, 1946, with the same dose to the brain and additional treatment to the spinal canal. In July, 1946, the patient began to suffer from pain in the arms and legs which was unusually severe.

Following the 3rd admission in July, 1946, roentgenograms of the skeleton showed widespread destruction of bone (Fig. 7), and a biopsy of a lesion in the right humerus was interpreted as showing neuroblastoma. Histological examination disclosed that

the bone was completely replaced by connective tissue containing large fibroblasts. The tumor cells were arranged in rosette shape. Most of the type cells were monopolar, some bipolar. Their nuclei were multiform, polygonal, oblong, often pear-shaped or triangular. The cytoplasm was confined to the prolongation or surrounded the nucleus in a small rim. The cells resembled those commonly described as neuroblasts (Fig. 8).

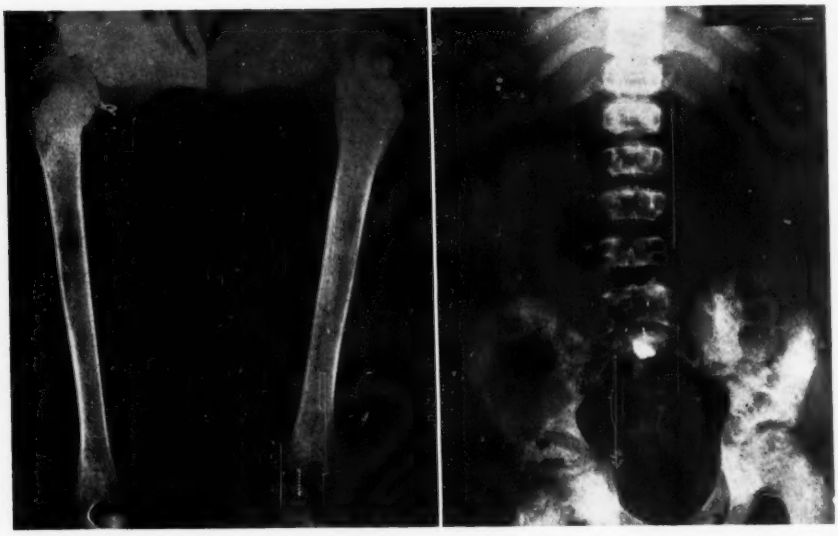

FIG. 7. Case 3. *Left*, Roentgenograms of each humerus of a 14-year-old boy previously treated for cerebellar medulloblastoma. Note irregular destruction and condensation of bone at the upper end of the diaphyses, and periosteal new bone formation on the right. *Right*, Spine and pelvis of same patient showing involvement of ribs, lumbar vertebrae, pelvis and hips. Biopsy from right humerus was reported as neuroblastoma.

Roentgen study of the urinary tract, abdomen, and chest showed no visible disease. The patient's bone pain was controlled by roentgen therapy. He left the hospital on Aug. 11, 1946, much improved but his condition deteriorated rapidly and he died a month later. No postmortem examination was obtained.

Comment. Here, again, a distinction between medulloblastoma and neuroblastoma could not be made on the sections from the original tumor, but the eventual outcome and later biopsy from the bone suggested the true nature of the disease.

DISCUSSION

If one turns to the classical original description of medulloblastoma from the pen of Harvey Cushing,[3] it is found that while most of the tumors he described occurred in children under 12 years of age, there were a few in older individuals in the 2nd and 3rd decades of life. Not only was the natura

history and prognosis more unpredictable in these few, but the position of the cerebellar tumors also differed from that in the younger group of patients. Although reluctant to eliminate these cases from his new classification, Cushing recognized their dissimilarity and suggested the possibility that some of them might be part of the group of neuroblastomas. Considerable argument has ensued since about this problem, and the opposing view-

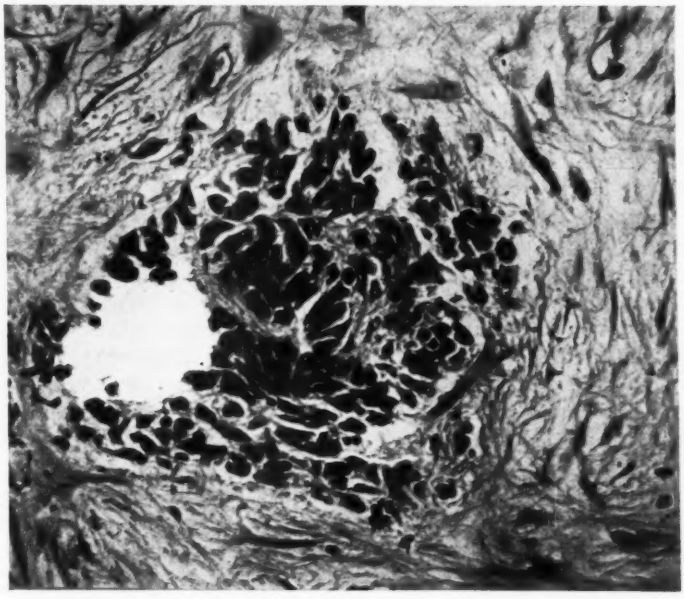

FIG. 8. Case 3. Bone tumor (neuroblastoma). Hematoxylin and eosin, ×440. Nest of tumor cells in completely destroyed bone whose tissue is replaced by connective tissue. Large fibroblasts are seen in this tissue. The tumor cells show rosette formation. The type cell is monopolar or bipolar. The multiform nuclei are surrounded by scant cytoplasm which may be confined to the prolongation. (Compare with Fig. 6.)

points of Penfield[11] and Stevenson[15] have been quoted to illustrate the uncertainty of the subject.

Our own experience indicates that there may be two types of medulloblastoma. The one is commonest in childhood and very malignant. Histologically, a diffuse arrangement of immature cells is prevalent in it. The other variant is more frequent in adults. It may be less malignant, with the patient surviving an unexpectedly long time.[6,12,14] Whorl and strand formations with reticulin septa are slightly more prevalent histologically in this group.[7] Widespread metastases to the rest of the body may develop, as illustrated by the 3 cases presented above. It is hoped, in a later essay, to present a basis for histologic differentiation between these two types of anaplastic cerebellar

tumors, so that with more accurate prognosis, a more selective therapy can be utilized.

If one considers the possibility that some of the cerebellar tumors in the older age group are neuroblastomas rather than medulloblastomas, the inconsistencies in the clinical course of these patients become explained. The natural history of abdominal neuroblastoma in patients over 12 years of age discloses the same two-fold potentiality. It is known that some of these tumors mature after radiation therapy, or even without any treatment, and become benign neurocytomas. Conversely, some of these patients present extensive metastases to the bones and viscera when they are first seen, and succumb rapidly. The protean manifestations of neuroblastoma in this age group are not widely appreciated and an understanding of them may help in evaluating some of the peculiarities of the cerebellar tumors under discussion.

Furthermore, it is possible that the cerebellar tumor for which a patient requires hospitalization is a metastasis from an occult primary tumor outside the central nervous system. Since it is conceivable that this situation may be present in any given case with the clinical and histological picture of medulloblastoma, it would seem essential that the pre-operative evaluation of the patient include a search for a primary tumor elsewhere than the brain, and particularly an intravenous urogram and roentgen survey of the skeleton and chest. If, as a result of these studies, the probability is that the cerebellar tumor is metastatic, a more conservative neurosurgical procedure may be indicated.

In addition to the difficulties of distinguishing between some cases of medulloblastoma and neuroblastoma, it should be pointed out that a similar confusion exists between neuroblastoma and Ewing's tumor.[2] Here, too, the clinical, radiological and pathological features of the two diseases may be so similar as to render differentiation between them extremely difficult.

Although thorough radiation therapy contributes a great deal to the management of the typical case of medulloblastoma,[4,9] it becomes of even greater importance in the group of atypical cases considered in this paper. While roentgen therapy is of value in palliation of most medulloblastomas, it may arrest the disease permanently in older patients with primary cerebellar tumors derived from neuroblasts, by influencing maturation of the cells in the same manner as in the abdominal neuroblastomas. Once metastases to the skeleton and chest have occurred, however, radiation therapy may relieve symptoms temporarily, but will not effect a cure.

CONCLUSION

1. Cerebellar tumors that are associated with similar tumors beyond the central nervous system are rare, but do occur.

2. Although presenting the histological picture of medulloblastoma, it is possible that they are in reality neuroblastomas.

3. Patients with clinical evidence of a cerebellar tumor, particularly

those in the 2nd and 3rd decades of life, should be subjected to pre-operative roentgenograms of the chest, skeleton and urinary tracts, to determine the presence of metastases or an occult primary tumor outside the central nervous system.

4. The difficulty of distinguishing some cases of cerebellar medulloblastoma from neuroblastoma by clinical, radiological or pathological methods parallels the situation in regard to neuroblastoma and Ewing's tumor.

5. Unexpectedly long survival of a patient with a cerebellar tumor resembling medulloblastoma, after thorough roentgen therapy, suggests that the tumor may have been a neuroblastoma which has undergone maturation and become arrested in growth.

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PROTRUDED LUMBAR INTERVERTEBRAL DISCS

RESULTS FOLLOWING SURGICAL AND NON-SURGICAL THERAPY*

B. M. SHINNERS, M.D., AND W. B. HAMBY, M.D.

Department of Neurology and Neurological Surgery, Buffalo General Hospital, and The University of Buffalo, School of Medicine and Dentistry, Buffalo, New York

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SINCE the original disclosure in 1934 by Mixter and Barr of the high incidence of protrusions of the lumbar intervertebral disc as a cause of low back pain and sciatica, many patients have been relieved of their complaints but new problems have arisen to perplex those dealing with these people. The growing population of persons who have been subjected to disc surgery has confronted practitioners and insurance adjustors with an increasing number of patients who have obtained unsatisfactory results. These focus unfavorable attention upon a group whose relative size, in proportion to the favorable group, is all too frequently ignored or misunderstood. It is helpful occasionally to demonstrate statistically the shifting proportions of these groups and several such reports have been published.[1,2,3,4,5,6,7,8] Fifteen years is too short a time in which to expect answers to all questions concerning a new clinical entity and its treatment, but evaluations of results obtained by various therapies is the means by which such answers can be developed.

Increasing attention is being given to the problem of the proper place or indication for spinal fusions after the removal of disc protrusions. At present, opinions range from that of Barr,[1] who feels that practically all backs should be fused after disc surgery, to that of Caldwell and Sheppard[2] who concluded that "there is no indication for spinal fusion" in patients having operations for disc protrusion.

We have attempted to find the answers to some of these questions in our own experience. It was not possible to re-examine personally a sufficiently large number of our patients, so we have conducted a second questionnaire survey of the patients we have treated surgically for this malady. A similar study was reported in 1944[5] of 140 patients who had been operated upon. In the present study, identical questionnaires were sent to 359 patients upon whom one of us (W.B.H.) had operated, including the original 140 reported previously. The data were collected and analyzed by one of us (B.M.S.), a neurologist.

THE MATERIAL

In dealing statistically with human material, the variables are so numerous that no criteria are entirely comparable in seemingly parallel series. Apparently similar disc protrusions compressing the same nerve roots do

* Read before the American Academy of Neurological Surgery, Montreal, September 21, 1948.

not produce uniform symptoms and signs nor are the same results obtained after removing the root-compressing protrusions. Results vary also with the type of work patients did before disability and to which they must return after operation. Varying compensation laws in different states may reward or actually place a financial penalty upon returning to a type of work that is optimal for the patient after operation. The age groups and the sex of patients vary in different series and alter the percentage results of surgery.

Sex. Of the 359 patients operated upon for back and leg pain, 113 were women and 246 were men.

Age. There were 46 (12.8 per cent) of our patients in the third decade (21–30 years) of life, 144 (40 per cent) in the fourth, 132 (36.8 per cent) in the fifth and 37 (10.3 per cent) in the sixth (51–60 years) decade. We have heard private expressions of opinion that patients older than 50 years do not develop disc protrusion; 10.3 per cent of our patients were in that age group.

Compensation. Of the 359 patients in this series, 237 (66 per cent) were "private" cases, who received no legal compensation for their injuries and 122 (34 per cent) were "compensation" cases.

Discs Involved. Of the 359 patients, 290 (81 per cent) had disc protrusions proved at a single level; 46 had multiple protrusions; in 23 patients re-operation was necessary because of recurrence of disc protrusion, 21 at the same level and 2 at a new level. We thus have found 392 instances of disc protrusion at various levels in the lumbar spinal canal.

Levels of Disc Protrusions. Fourteen disc protrusions not included in these statistics were found located at the C3 level in 1 case, at C4 in 1 case, C5 in 4 cases, C6 in 4 cases, C7 in 1 case, D8 in 1 case and D10 in 2 cases. The lumbar disc protrusions were found at L3 in 14 cases (4 on the left side, 3 midline and 7 on the right), L4 in 155 cases (93 left, 8 midline, and 54 right) and at L5 in 223 cases (107 left, 17 midline and 99 right). The 392 lumbar disc protrusions were found on the left side in 204 cases, midline in 28 and on the right in 160 cases. There did not seem to be any pertinent correlation with the dominant "handedness" of the patients.

Follow-up Period. The longest follow-up period after operation was 11 years; the shortest, 1 year and the average was 4.3 years. The average follow-up period for patients with spinal fusion was 3.0 years.

RESULTS FOLLOWING SURGERY

Questionnaires (Table 1) were sent to 359 patients who had been operated upon: 300 patients responded promptly and over the course of a year; with the assistance of interested referring physicians and insurance adjustors, 355 (98.7 per cent) responses were obtained.

The Operations. In 289 (81.5 per cent) cases, the operation had consisted of removal of the disc protrusion by approach between the laminas in the vast majority of cases, by hemilaminectomy in a few of the early cases and by laminectomy in a very few of the complicated cases. The protrusion itself was treated by as simple removal as possible, but the sinus leading

into the disc was curetted of all easily removable fragments. Since they have been available, fibrin foam or gelatin sponge have been left in the epidural space for control of epidural venous bleeding.

In 66 cases (18.5 per cent) the involved segments of the spine were fused

TABLE 1

QUESTIONNAIRE SENT TO PATIENTS OPERATED UPON

140 Linwood Ave.
Buffalo 9, N. Y.

Dear Sir:

I am interested in finding out the results of the operations performed on some of my former patients. With this in mind, I am sending you this mimeographed letter, hoping that you will answer the questions and return it to me.

1. PLEASE PUT AN *X* IN FRONT OF ONE OF THESE STATEMENTS:
   I think the operation cured me.
   I think the operation helped, but did not cure me.
   I think I am about the same as before the operation.
   I think I am worse than I was before the operation.
2. Did you feel relieved of pain before you left the hospital? _____
3. When did you begin feeling better? _____
4. Do you still have pain? _____ If so, describe what it is like. _____
   _____
5. Are you working? _____ When did you return to work? _____
   (a) What kind of work are you doing? _____
   (b) What kind of work did you do before the operation? _____
   _____
   (c) What kind of work did you do before your trouble started? _____
   _____
   (d) Are you getting compensation now? _____
   (e) Does working bother your back? _____ Your legs? _____
6. Are you worse in any way than before the operation? _____
   If so, please tell how _____
7. If you are not well, are you in any way better than you were before the operation? _____ Please tell how _____
8. In general, are you glad or sorry you had the operation? _____
   _____

Thank you for your help.

Very truly yours,

by an orthopedist for one indication or another. The fusion was done immediately after the disc operation in 49 cases. In the other 17 cases the fusion was done some time later because of persistence of symptoms after the disc operation.

TABLE 2

The personal evaluation of their status following operation, by 354 patients operated upon for protruded intervertebral discs

	Removal Disc Protrusions	Plus Spinal Fusion	Totals
Cured	150 (52.0%)	22 (33.3%)	172 (48.5%)
Helped	107 (37.2%)	31 (46.9%)	138 (39.0%)
No change	28 (9.7%)	11 (16.6%)	39 (11.0%)
Worse	3 (1.1%)	2 (3.3%)	5 (1.5%)
Totals	288 (100.0%)	66 (100.0%)	354 (100.0%)

The patients' personal evaluations of their postoperative status are summarized in Table 2.

Of the 289 patients who had the simple disc operation, 192 (66.5 per cent) reported that they were relieved of their pain immediately after operation and 200 (69.3 per cent) were free of pain before leaving the hospital. At the time of the survey the pain-free group had declined to 47 per cent. On this service the patients usually are discharged from the hospital on the 14th postoperative day.

Responses to the 4th and 5th questions are summarized in Table 3.

The responses to the factor of compensation are summarized in Table 4.

Of the 355 patients operated upon, 235 (66.2 per cent) were "private" cases and 120 (33.8 per cent) were "compensation" cases. A comparison of the results obtained in the two groups is interesting to the insurance carriers as well as to physicians. Of the private patients, 142 (59.5 per cent) considered themselves well, while only 35 (29 per cent) of the 120 compensation cases would rate themselves so. There were 75 (32 per cent) private patients and 60 (50 per cent) of the 120 compensation patients who admitted being "helped" or improved by the operation. This is a source of annoyance to the insurance carriers, since it prevents them from clearing the cases from their books. This contributes to their growing reluctance to approve of disc sur-

TABLE 3

Postoperative status of 355 patients as to pain and ability to work

	Simple Operation	Plus Fusion	Totals
No pain	140 (48.5%)	27 (40.9%)	167 (47.0%)
Back pain	120 (41.5%)	24 (36.4%)	144 (40.6%)
Leg pain	29 (10.0%)	15 (22.7%)	44 (12.4%)
Totals	289 (100.0%)	66 (100.0%)	355 (100.0%)
Are working	271 (93.8%)	54 (81.8%)	325 (91.5%)
Are not working	18 (6.2%)	12 (18.2%)	30 (08.5%)
Totals	289 (100.0%)	66 (100.0%)	355 (100.0%)

gery. From the patients' standpoint the difference is very real. The private patient works in spite of, or adjusts his work to some postoperative disability, but the compensation patient who admits that he is cured must expect to return to his original work, which may be too heavy for his back after operation. In our 1944 survey little difference was noted in the results admitted by the two groups of patients. It seems likely that in the interval, from contact with increasing numbers of fellow workers who have been operated upon, the compensation patient is becoming more reluctant to admit being "cured" by surgery. Since the selection of patients and the technic of opera-

TABLE 4

Contrasts in results obtained by disc operation with and without spinal fusion in 355 patients, 236 (66.2%) of whom were "private" and 120 (33.8%) "compensation" cases

	Disc Operation		Plus Fusion		Totals
	Private	Compensation	Private	Compensation	
Cured	127 (62.7%)	27 (31.0%)	15 (45.5%)	8 (24.2%)	177 (50.0%)
Helped	60 (30.0%)	42 (48.2%)	15 (45.5%)	18 (54.5%)	135 (38.0%)
No change	11 (5.4%)	16 (18.4%)	3 (9.0%)	5 (15.1%)	35 (9.7%)
Worse	4 (1.9%)	2 (2.4%)		2 (6.2%)	8 (2.3%)
Totals	202 (100.0%)	87 (100.0%)	33 (100.0%)	33 (100.0%)	355 (100.0%)
	33	33			
	235 (66.2%)	120 (33.8%) = 355 (100%)			

tion have not changed appreciably, it is not likely that actual surgical results have changed. Indeed, the rates of "cures" in the private cases have improved from 53.3 per cent in the 1944 report to 62.7 per cent, while the rate in compensation cases has fallen by 10 per cent. It appears then, that the difficulty is not in the therapy, but in the rules for compensation adjustment.

FATE OF UNOPERATED CASES

Apropos of the compensation dilemma, it would be most helpful if some insurance company would statistically survey its groups of patients disabled by low back pain and sciatica (including such classifications as "sacro-iliac strain," "lumbago," etc.) for equal time periods before and since the advent of the surgical treatment of disc protrusions. We know of no report of such a survey. A similar study of patients seen by any physician or clinic would be too heavily weighted to be as significant statistically. In an attempt to evaluate our own experience in this respect, we took from our files records of 1000 patients who had been seen because of low back pain and sciatica and who had not been operated upon. From the records, 650 of these seemed likely to have had disc protrusions, but only 300 gave histories and had objective neurologic findings that we considered significant of nerve root compression. To these 300 selected patients another questionnaire (Table 5) was sent.

Of these, 225 patients (75 per cent) answered this inquiry; 25 of them had been operated upon elsewhere, so were eliminated, leaving 200 whose data were analyzed.

Results of Survey. The average follow-up period was approximately the same as in the surgical group. Of the 200 patients, 59 (29.5 per cent) considered themselves well, in contrast to 172 (48.5 per cent) of the 354 patients who were operated upon; 141 (70 per cent) of the 200 continue to have quite

TABLE 5

QUESTIONNAIRE SENT TO PATIENTS NOT OPERATED UPON

140 Linwood Avenue
Buffalo 9, New York

Dear Sir:

We are attempting to find out the effect of various types of treatment of people who have had back or leg pain.

On _____ I had the opportunity of examining you for back or leg pain. I am taking the privilege of sending this mimeographed letter to determine whether you are well or have had further trouble with your back or leg.

I hope you will fill this form out and return it to me. (The information will be confidential.)

1. Are you still having back pain? Yes ☐ No ☐
2. Are you still having leg pain? Yes ☐ No ☐
3. Are you still under a doctor's care for either back or leg pain?

   Yes ☐ No ☐
4. Approximately how many attacks of leg pain have you had since your examination here?

   _____
5. Have you had an operation for your back or leg pain? _____
6. Have you changed your work because of your pain? _____
7. Are you doing the same type of work? Yes ☐ No ☐
8. What type of work are you doing now? _____

Please add any extra comment that you might have on the back of this letter. We thank you in advance.

Very truly yours,

constant back pain. More than 50 per cent have recurrent attacks of back and leg pain; 63 per cent of these 200 people continue seeing physicians periodically and the remaining 7.5 per cent of them have become resigned to their fate or rely upon the ministrations of cultists.

Work Record. More patients in both the surgical and the non-surgical groups are working than admit being well. Of the patients treated surgically, 91.6 per cent are working, whether they had the simple disc operation or that combined with spinal fusion. Of the patients not operated upon, 86 per cent are able to work. Approximately 20 per cent of the surgically treated group and 25 per cent of those not operated upon have found it necessary to change to lighter work. As shown earlier (Table 3), 142 (60 per cent) of

the 235 private patients in the surgical group consider themselves cured, while only 35 (29 per cent) of the 120 compensation patients consider themselves cured by operation. Of the 200 non-surgical cases, 31 were compensation patients; 16 (51.5 per cent) are not working. The evidence from these two series certainly does not indicate that the pessimism of the insurance carriers is justified.

ADDITIONAL DISCUSSION

While complete statistical analysis has not been made in all points to be discussed here, several pertinent conclusions concerning the disc protrusion problem seemed evident.

Patients who displayed objective neurologic evidence of nerve root compression obtained definitely greater benefit from surgical removal of the disc protrusion than those who did not. Patients with constant backache and only occasional twinges of leg pain, without objective neurologic findings, had poor results from disc surgery. Patients who required spinograms for diagnosis (not for localization) of their lesions were less often helped by operation than those with objective evidence of nerve root compression. Patients with severe sciatica before operation are less likely to be concerned by residual backache after the protrusion is removed. Patients operated upon in the acute phase of the illness obtained better results than those operated upon in a relatively pain-free remission. The best results were obtained from decompression of nerve roots by simple removal of disc protrusions.

Spinal fusion done after the removal of disc protrusions does not prevent postoperative backache, obviate the necessity for subsequent operations, nor prevent recurrence of disc protrusion. In our series a higher percentage of back and leg pain followed spinal fusion than after simple removal of the disc protrusion. This is in accord with the observations of Smith[6] based on more than 10 years of familiarity with disability awards by the Oregon State Industrial Accident Commission. He said, "Experience with partial hemilaminectomy for protruded intervertebral disc has, on the whole, been very satisfactory." And "A study of our records reveals that in cases in which the combined operation (disc protrusion removal and spinal fusion) has been carried out, the permanent disability averages 40 per cent greater than in cases in which hemilaminectomy alone has been done." The patients who were operated upon, both those with and without fusion, however, showed marked improvement when contrasted with the patients not operated upon.

SUMMARY

Questionnaire surveys were made of the results obtained in the surgical treatment of 355 patients having lumbar intervertebral disc protrusions and of the present status of 200 such patients who were not operated upon.

Of the surgical group 48.5 per cent considered themselves cured and 39 per cent were benefited by operation, while only 29.5 per cent of the unoperated group had recovered.

There are 91.5 per cent of the surgical group and 86 per cent of the unoperated group working. Only 50 per cent of unoperated compensation patients are working.

Simple removal of disc protrusions was done in 81 per cent of the surgical patients. This was combined or followed by spinal fusion in 19 per cent.

Simple removal of disc protrusions was followed by definitely better results than when spinal fusion also was done.

In contrast to results obtained in a similar previous survey, definitely better surgical results were obtained with private than with compensation patients; probable reasons are suggested for this.

Other factors are discussed relative to improving results of intervertebral disc surgery.

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CHRONIC POSTTRAUMATIC HEADACHE

AND THE SYNDROME OF CERVICAL DISC LESION FOLLOWING HEAD TRAUMA

AIDAN A. RANEY, M.D., R. B. RANEY, M.D., AND C. R. HUNTER, M.D.
*Los Angeles, California**

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PUBLISHED reports indicate considerable variance in opinion on the subject of etiology of chronic posttraumatic headache, although it is generally attributed to an intracranial process. There is accumulated evidence that cervical intervertebral disc pathology is often responsible. Therefore, it seems appropriate to survey pertinent clinical and experimental observations related to this interesting subject.

THEORIES ON ETIOLOGY

A. *Intracranial Structures.* The effect of trauma of the intracranial structures is often assumed to be responsible for chronic posttraumatic headaches. Yet, there is substantial evidence opposed to this theory.

The trauma of neurosurgical procedures that does not affect the cervical region but does cause extensive cranial, meningeal, and cerebral defects, is not often followed by chronic headache. On this account it seems illogical to conclude that disturbance of continuity of these structures or meningeal adhesions is responsible for the headache. On the other hand, clinical observations show that trauma may be an important etiologic factor in chronic headache.

Nielsen,[28] Symonds,[47] Penfield and Shaver,[33] and others have pointed out that many patients with severe head injury make prompt and complete clinical recovery; in other cases of slight head trauma with transitory or no loss of consciousness, persistent and disabling posttraumatic headache ensues. For example, Coleman[2] observed, among 19 patients rendered unconscious by trauma, 16 who complained of troublesome headaches. On the other hand, in 8 cases of head injury without loss of consciousness, 7 had annoying posttraumatic headaches. Glaser and Shafer[9] observed that posttraumatic symptoms, of which headache was the most frequent, were not proportionate to the duration of unconsciousness. Symonds[47] stated that posttraumatic symptoms could not be regarded merely as a sequel of the general effect of brain injury; furthermore, such symptoms were not, as a rule, associated with evidence of brain injury or focal brain damage.

Russell[41] found that headache was severe in most cases of high intracranial pressure due to acute trauma. Yet, in some cases of equally high pressure there was no complaint of this symptom. Others[8,38,51] have found no

* 1136 West Sixth Street, Los Angeles 14, California.

convincing evidence that the increase of intracranial pressure in itself is responsible for subsequent posttraumatic headache.

Friedman and Merritt,[8] in a study of 265 cases of head trauma, found that chronic posttraumatic headache occurred with equal frequency in those who had bloody cerebrospinal fluid and those with clear fluid following injury. Penfield and Shaver[33] believed that bloody spinal fluid was not an important etiologic factor in such headaches. A definite relationship has not been established between the concentration of blood in the cerebrospinal fluid and subsequent chronic headache.

Troland, Baxter and Schatzki[48] found, in 116 patients with traumatic cranial defects, that only 41 per cent had posttraumatic headache, dizziness, and so forth. This is particularly significant because, since the cranial defects in their cases were due to war injuries, it seems fair to assume the majority had meningeal involvement. Courville[3] states that there are no known gross alterations in the brain or meninges to account for headache and certain other symptoms of the postconcussion syndrome. These observations tend to discredit the theory of subdural traction[32] and involvement of the meninges[30] as causes for chronic headache.

On the basis of reports, there seems to be no justification for routinely ascribing chronic posttraumatic headache to the effects of trauma on the intracranial structures. However, since trauma is a factor present in each instance, its relationship to subsequent headache cannot be denied. Its effects on other structures are worthy of attention.

B. *The Skull.* Glaser and Shafer[9] found that 90 per cent of their patients with head injury without skull fracture complained of symptoms, yet only 40 per cent of those with depressed vault fracture complained of symptoms. Penfield and Shaver[33] noted a higher incidence of headache in patients with fractures of the skull, but their observations did not lead them to conclude that the fracture was significantly related to the headache.

C. *The Scalp.* Because traumatic lesions of the skull and its contents have failed to explain posttraumatic headache, extracranial structures have been studied. Watts, Wiley, and Groh[50] believed that headache, and dizziness also, following head injury was often due to contusion of the scalp. They observed that posttraumatic headache was frequently relieved by infiltration of procaine around the artery at the site of pain and they theorized that this finding was an indication that the posttraumatic headache was in reality a syndrome of vascular pain (as described by Fay[6]) and similar in mechanism to migraine headache (as described by Graham and Wolff[10]).

Stimulated by encouraging reports on the use of prostigmine[26] and histamine,[7] Robertson and Peacher[39] used these drugs alternately in cases of posttraumatic headache. They found neither drug sufficiently effective in 50 cases to justify continuation of the series.

The high percentage of cases in which extensive traumatic or surgical lesions of the scalp and larger arteries are followed by complete symptomatic recovery likewise does not support the theory of headache being due to

scalp injury. For these and other reasons, it is believed that neither scalp injury nor the vascular theory alone is generally acceptable in explanation of chronic posttraumatic headache.

D. *Cervical Sources of Headache.* For almost 200 years reports have appeared in support of a cervical origin of headache.[12] As previously pointed out,[36] arthritis, myositis, spondylitis, tumors, Pott's disease and similar lesions in the neck have been reported as causes for head pain. In 1913 Luff[24] stated that fibrositis in the neck sometimes caused headache. More recently Kelly[18] and others have emphasized the importance of such lesions as sources of head pain. We have observed that simple irritation of the periosteum of the occipital plate by the point of a needle causes pain often referred to a circumscribed area in the frontal or temporal region. The point to which the pain is referred shifts in a predictable pattern as the point of the needle is moved to a new location.

By injecting hypertonic saline into the suboccipital attachment of muscles, Cyriax[4] produced pain spreading around the head to the temple and forehead. Kellgren[16] called attention to referred pain of muscle origin and its relief by procaine infiltration. Kelly,[18] stimulated by these reports, made extensive use of procaine infiltration in investigation and treatment of head pain, including posttraumatic headache, which he attributed to fibrositis of the cervical muscles. He stated that fibrositic lesions above the 4th cervical vertebra may cause headache, and if the lesion is deeply situated in the suboccipital region *there may be no local pain, all the pain being referred to the frontal or orbital region.* He believed fibrositis was responsible for many cases of posttraumatic headache.

Jones and Brown,[14] believing that posttraumatic head pain may be caused by traumatic fibrositis of the structures surrounding the neurovascular bundle, treated 120 patients by procaine injection. In 50 per cent they obtained good results, the patients being relieved completely or sufficiently to permit return to their regular occupations. Young[52] observed that pain originating in the neck may radiate to the eye, although he expressed some doubt that fibrositis was a primary pathologic entity. Campbell and Parsons,[1] observing that head pain may be referred from other regions, listed lesions of muscle, bone, fascia, and nerve tissue in the neck as potential sources. In an analysis of 1000 cases of headache, Hersh[12] found 24 per cent due to myositis cervicalis. In 8 cases arthritic changes were noted, and in 1 case disc herniation was diagnosed, although he observes that scant attention has been directed to these conditions. Jackson[13] has reported on lesions in the cervical region causing head pain.

In view of these experimental and clinical observations we believe it reasonable to accept as fact that headache can, and frequently does, originate from a pathologic process in the cervical region. Therefore, the neck should be investigated in cases of posttraumatic headache of undetermined origin.

INJURY OF THE NECK RESULTING FROM HEAD TRAUMA

In almost every instance of head trauma, some of the force is transmitted to the structures of the neck. If the trauma is severe enough to cause skull fracture or loss of consciousness, it is more probable that secondary trauma has caused serious damage to the structures of the neck. Notable exceptions are those cases of head injury caused by small missiles of high velocity. In these cases the inertia of the head serves to prevent associated neck injury. This opinion is supported by clinical observations of Robertson and Peacher,[39] who state that the manifestations of the posttraumatic syndrome are not often observed after the more severe penetrating gunshot wounds.

Meredith and Grantham[27] reported 6 cases in which fracture or dislocation of the cervical spine resulted from simultaneous direct trauma of the head and shoulder (Coleman's syndrome). Walshe[49] called attention to combined head and neck injury with minor cord involvement which may be overlooked or attributed to a brain injury. In a discussion of headaches associated with injuries of the cervical spine, Little[23] reports a case in which a cervical disc lesion was responsible for headache. Thus, it is apparent that any investigation of the cervical region for effects of trauma which terminates with a roentgenographic report that no fracture, dislocation, or bony pathology is noted, may overlook important lesions capable of causing chronic and disabling symptoms.

LESION OF CERVICAL DISC

Headache is often a symptom of cervical disc lesion in the early stages of pathology preceding the development of radicular symptoms. Headache may be regarded as the "lumbago" of cervical disc lesions, corresponding to "lumbago" of the low back which usually precedes the radicular symptoms of lumbar disc herniation.[34,36]

More recently Jackson,[13] in her report on 200 patients with cervical spine lesions causing shoulder disability, states that in 100 patients headache was one of the principal symptoms and 35 per cent had roentgenographic evidence of ruptured cervical disc.

Development or exacerbation of symptoms of disc origin commonly results from trauma, even though the trauma is slight. Kristoff and Odom[20] believe that probably all ruptures of cervical discs are traumatic in origin. Disc disease is of such frequency that its symptoms often appear following trauma transmitted to the cervical spine from head injury. Head trauma is, of course, not the sole etiologic factor in headache of cervical disc origin. Pathology of a cervical disc may be produced by, or become symptomatic following, direct trauma to the neck, torsion or other strains of the cervical spine. For example, acute flexion of the cervical spine during the administration of spinal anesthesia may cause postspinal headache.[34] The amount of force necessary to initiate or cause exacerbation of symptoms due to disc disease is dependent upon the presence or degree of preexisting disc disease

and the protection against injury afforded by muscle guarding and the postural attitude of the cervical spine at the time of trauma. Other significant features of this clinical entity have been discussed in our previous reports.[34,35,36]

The interior of the disc is devoid of nerve tissue[15] and, therefore, the degeneration itself is not the immediate cause of the symptoms. The degeneration does, however, predispose to effects on local sensitive structures. Mechanical factors frequently precipitate these effects on adjacent tissues, resulting in local pain or muscle spasm and producing pain localized in the muscle and its attachments or pain referred to other regions. Thus, pain of disc origin can be expected to vary in intensity, frequency and duration. The most important single feature of the pain caused by this physical derangement is that it is influenced by mechanical factors. Pressure on the vertex of the head, for example, may cause or increase pain of cervical disc origin,[45] and the manual head traction test[36] often affords temporary relief from headache or other pain of cervical disc origin. These are important diagnostic signs. Infiltration anesthesia of the spastic (fibrositic?) muscles and the site of reference of the pain often affords relief.

Natural reparative processes, so effective in rendering asymptomatic such lesions as lacerations, fractures, muscle injury, and so forth, are retarded in disc lesions because of the nature of the affected tissue. Its avascularity prevents invasion of fibroblasts (and intrinsic repair of the disc lesion) as occurs in lesions of other tissues. Consequently, the symptoms of a defective disc may continue until nature is able to render the pathologic process asymptomatic by the indirect method of external fibrous and arthritic immobilization of the joint. Increased muscle tension in symptomatic disc lesions may produce a reduction or loss of the normal lordotic curvature of the cervical spine. It is significant that this characteristic attitude is maintained permanently when the lesion finally heals by "segmental hypertrophic arthritis." That primary lesions of the intervertebral disc are a common cause of exostotic formations known as hypertrophic arthritis, deforming spondylitis, etc., has been shown.[19,29,43]

DISCUSSION

Pain directly due to painful stimulation of the structures innervated by the trigeminal nerve is usually confined to the trigeminal area. This has been shown by experimental stimulation of the eye,[5] nasal and paranasal structures,[25] teeth,[40] and the intracranial structures as far posterior as the superior surface of the tentorium.[31] The reference of pain to the frontal or orbital region from stimulation of intracranial supratentorial structures is logical because they are innervated by a branch from the ophthalmic division of the trigeminal nerve.[38] Excessive and sustained painful stimulation of structures in the trigeminal area may produce spasm of the nuchal muscles and pain may then develop in the neck, such pain being a secondary effect and not referred pain from the site of primary stimulation.[25,40,44] Stimulation of

structures previously rendered anesthetic by trigeminal root section produced neither local pain nor pain elsewhere.[25,40]

Kellgren[17] found that pain resulting from local stimulation of muscle bellies is felt diffusely over a considerable area which may be situated at a distance from the point stimulated. For example, hypertonic saline injection of the small suboccipital muscles produces deeply situated headache. The pain commonly referred to as headache is an "aching" pain and similar to that described by Lewis[21,22] as arising from muscle and other deep somatic structures.

Campbell and Parsons[1] state that the evidence of intracranial causes of many types of headache, varying from postconcussive to migraine, is inferential or non-existent. They regard bony, fascial, muscular, and nerve lesions of the occipito-cervical region as potential causes for the headache. By injection of hypertonic saline in the occipital and upper cervical region, they produced pain not only locally but, more significantly, pain referred to the top, sides, and even the front of the head. These injections almost constantly produced giddiness and listing, and such autonomic disturbances as pallor, sweating, nausea and alterations of the pulse were frequently observed. They observed that this experimentally produced state bore a marked resemblance to the posttraumatic syndrome. Stockman[46] and Gutstein-Good[11] believed vertigo to be commonly a part of the syndrome of cervical lesions. These studies and those of Cyriax[4] demonstrate that headache even in the frontal region may be due to a pathologic process in the suboccipital or cervical region. Kelly,[18] Jones and Brown[14] and others made similar observations.

Several observers have attributed chronic symptoms and findings to fibrositis, assuming it to be a primary pathologic entity. However, microscopic studies of such so-called fibrositic lesions have shown no pathologic changes.[42] Many features of the clinical signs, particularly chronicity and recurrence, are difficult to explain on the theory that fibrositis is the primary lesion. Such features are more logically explained if these soft tissue reactions are regarded as processes secondary to a pathologic disc or other lesion in the neck.

Perhaps pain in the frontal region, for example, due to injection of irritants in the suboccipital area, may at times be explainable on the basis of spasm of the frontalis muscle secondary to traction on the galea from occipitalis spasm. Sometimes patients complain of frontal headache and subsequently realize that the diffuse deep-seated pain is actually occipital or suboccipital in location. Perhaps there are additional explanations[37] for discrepancy between the location of the pathology and the patient's localization of the discomfort that it produces.

SUMMARY AND CONCLUSIONS

Chronic posttraumatic headache is often unexplainable on the basis of lesions of the scalp, skull or intracranial contents. The posttraumatic syn-

drome has been duplicated experimentally by injection of irritating solutions into the upper cervical and suboccipital regions. There is clinical evidence that chronic posttraumatic headache may arise from lesions in the neck. We believe that disc lesions often are the underlying pathology responsible for such processes as segmental hypertrophic cervical arthritis, myositis, and fibrositis—conditions to which headache has been attributed.

The intervertebral disc is avascular and, therefore, slow in healing. Lesion of a cervical disc, therefore, fulfills the requirements as a cause for chronic and recurrent symptoms more completely than do lesions of tissues possessing a better blood supply.

The foregoing data indicate that a search for potential sources of chronic posttraumatic headache is incomplete if it fails to include investigation for evidence of cervical disc defects.

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TRANSIENT HEMIPLEGIA ASSOCIATED WITH CEREBRAL ANGIOGRAPHY (DIODRAST)*

J. G. CHUSID, M.D., FRANKLIN ROBINSON, M.D.,
AND M. P. MARGULES-LAVERGNE, M.D.
The Neurological Division, St. Vincent's Hospital, New York City

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SINCE the introduction of cerebral angiography by Egas Moniz,[20] repeated efforts have been made toward the simplification of the technique as well as the reduction in hazard for the patient undergoing the diagnostic procedure. The wider adoption of the percutaneous injection method of the carotid system[4,6,16,18,23] and the use of seriographic devices[26,27] have overcome some of the difficulties formerly encountered. However, the development of an inert substance, ideal for radiographic visualization, is still forthcoming. Thorotrast is considered by some investigators[12,17] superior to diodrast because of its relatively greater radiopacity and less frequent immediate systemic reactions. Nevertheless, because of the potentially dangerous radioactivity of thorotrast, other workers[6,7,8,9,10,14,16,22,23] have employed diodrast as the contrast medium of choice.

Recently, the authors have observed the occurrence of transient hemiplegia in 2 patients as an immediate complication of diodrast cerebral angiography. A review of the literature failed to disclose reports of such transitory hemiplegic phenomena with the use of diodrast in concentration of 35 per cent. These cases are presented, therefore, together with a brief résumé of the pharmacological action of diodrast and the clinical experience of others with its use.

Case 1. M. C., a 49-year-old white male, was admitted to St. Vincent's Hospital on 25 Oct. 1948, 10 hours following the onset of severe supraorbital headache, vertigo, nausea and projectile vomiting. Neurological findings were normal except for moderate stiffness of the neck. The patient was right-handed. Blood pressure was 140/100 and temperature, pulse and respiration were normal. Lumbar puncture disclosed grossly bloody fluid with an initial pressure of 290 mm. By the 12th day the CSF was no longer bloody and the initial pressure was 156 mm. Gradual symptomatic improvement occurred during the next 2 weeks. Laboratory studies including complete blood count, bleeding and coagulation time, blood and spinal fluid serology, visual fields and skull x-rays were within normal limits. An EEG (Fig. 1) on the 17th day was borderline. The patient denied any personal or familial allergic reactions to food, pollen or bacteria. An ocular test[1] for diodrast sensitivity was negative.

On the 32nd day *right percutaneous cerebral angiography* was performed under local anesthesia, using a No. 18 needle. Premedication consisted of nembutal 0.10

* Presented before the New York Society of Neurosurgery, at the New York Academy of Medicine, 15 March 1949. In the general discussion, 21 additional similar complications following cerebral angiography with 35 per cent diodrast were reported.

gm. and codeine phosphate 0.06 gm. The right common carotid artery was entered without difficulty and a right lateral projection arteriogram and venogram was obtained with 20 cc. of 35 per cent diodrast. There was incomplete detail in the course of the right anterior cerebral artery and a repeat injection was performed using an equal amount of diodrast. Thirty seconds following the injection the patient lost consciousness and remained unresponsive for approximately 2 minutes. No convulsive movements were observed. A complete left flaccid hemiplegia with weakness of the left lower facial musculature was evident within 3 minutes. The deep reflexes on the left side were increased slightly, the left plantar response was

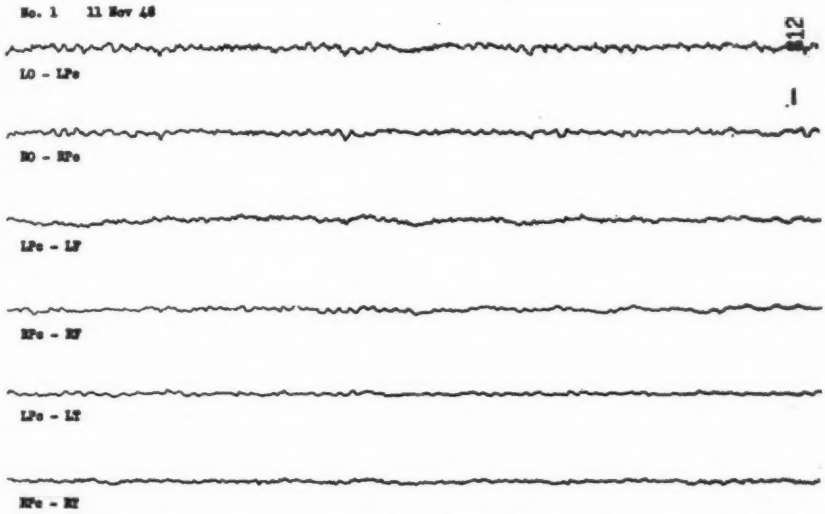

FIG. 1. Record made 2½ weeks after subarachnoid hemorrhage and prior to cerebral angiography, reported as borderline because of prominent 6 to 8/sec. waves (Case 1).
L=Left R=Right F=Frontal Pc=Precentral T=Temporal O=Occipital
Calibration same for all records (50 microvolts and 1 second).

extensor, and there was left ankle clonus. The blood pressure was recorded at 160/100 and pulse rate 76. The patient was given 300 mg. of tetraethylammonium chloride intravenously approximately 10 minutes later with immediate fall in blood pressure to 106/90 and rise in pulse rate to 96. The patient remained lethargic and moderately dysphasic, but return in motor function of the extremities of the left side occurred within 1½ hours after onset of the episode. At that time, the deep reflexes were equal and active bilaterally and the left plantar response was flexor. Another intravenous injection of 200 mg. of tetraethylammonium chloride was given. Approximately 2½ hours after onset of the reaction, the patient was alert, orientated, and there was no evidence of speech disturbance.

On the 37th day *left cerebral angiography* was performed using the same technique. The procedure was tolerated well and there was no discomfort. A small aneurysmal dilation of the left anterior cerebral artery just distal to the anterior communicating artery was disclosed. The remainder of the patient's course was uneventful and he was discharged on the 44th hospital day. He remained asympto-

matic and neurological findings on 8 Mar. 1949 were within normal limits. An EEG tracing (Fig. 2) made on 20 Jan. 1949 had a normal pattern.

Case 2. W. T., a 49-year-old white married carpenter, was admitted to St. Vincent's Hospital on 4 Jan. 1949 for study. He had had an episode of severe left-sided headache associated with nausea and vomiting a year ago with apparent complete recovery. He remained well until the morning of 7 Dec. 1948, when, while bending over, severe generalized headache developed, radiating into the back of the neck. The headache persisted, and on the following day he was admitted to a

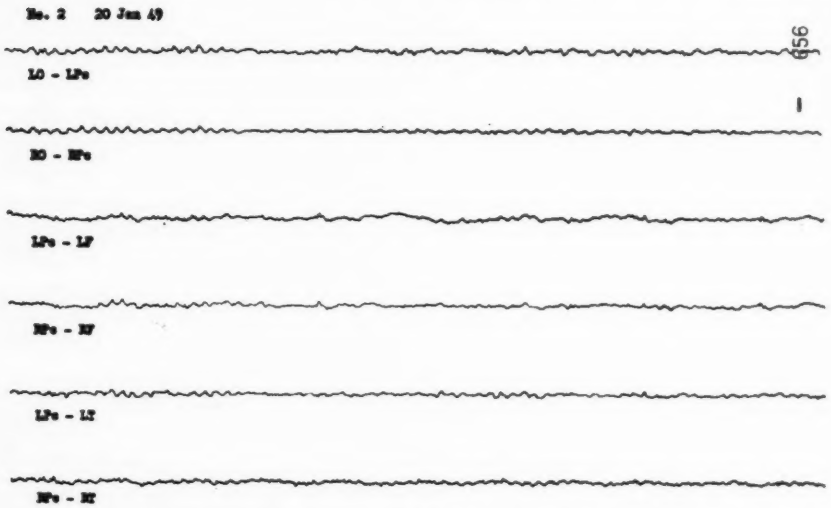

FIG. 2. Normal EEG made approximately 2 months after transient left hemiplegia associated with right cerebral angiography (Case 1).

hospital. Initial lumbar puncture revealed grossly bloody fluid under 180 mm. pressure. There was nystagmus on right lateral gaze with absence of right abdominal reflexes. Subsequent punctures revealed progressive clearing of the CSF with diminishing pressure, and he was asymptomatic on discharge. Neurological findings on admission to St. Vincent's Hospital were within normal limits. The patient was right-handed. Studies, including skull x-ray, EEG (Fig. 3), complete blood count, urinalysis, blood urea nitrogen, fasting blood sugar and blood Kahn, were not remarkable. The patient and his family denied sensitivity to food, pollen or bacteria. An ocular test[1] for diodrast sensitivity was negative.

Left percutaneous cerebral angiography was performed under local anesthesia on the 2nd hospital day. Two injections of 35 per cent diodrast were made, using a total of 35 cc. of solution for the arterial and venous phases in the left lateral projection with no untoward effect. A moderately large saccular aneurysm of the intracranial portion of the left internal carotid artery was seen. A 3rd injection of 15 cc. of 35 per cent diodrast was made in order to obtain an A-P projection. Promptly following this injection he became confused and dysphasic. A right flaccid hemiparesis with inactive right plantar response was evident. The blood pressure

was 108/60. The patient received an infusion of 500 cc. of saline containing 500 mg. of tetraethylammonium chloride and appeared less confused. Two hours later full power of the right lower extremity was regained and the right plantar response was flexor. Flaccid paralysis of the right upper extremity and the lower right side of the face persisted. Lumbar puncture, 3 hours after onset of the reaction, yielded clear fluid under initial pressure of 110 mm., with 6 cells / c.mm., total protein 38 mg. per cent, and colloidal gold of 1122110000. Motor power in the right upper extremity continued to improve and 4 days later, motor function and speech were normal.

On the 22nd hospital day left carotid compression, starting with a 5-minute

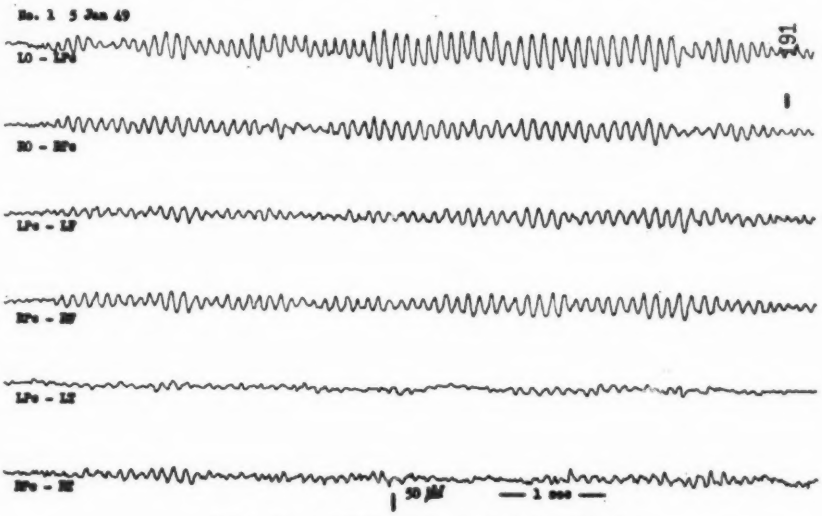

FIG. 3. Normal EEG present on admission (Case 2).

period, was begun without ill effect. By the 27th day, the patient tolerated 20 minutes of carotid compression quite well. On 1 Feb. 1949, the 28th hospital day, the left common carotid was exposed and ligated under local anesthesia, with uneventful postoperative course. On the 6th postoperative day *right cerebral angiography* was performed under local anesthesia. A total of 40 cc. of 35 per cent diodrast was used without side effect. The vascular pattern appeared normal and the aneurysm of the left side did not fill. The patient was discharged on the 9th postoperative day.

The initial EEG (Fig. 3) taken on 5 Jan. 1949 was normal. The 2nd EEG (Fig. 4) 7 Jan. 1949, taken 1 day following the right hemiplegia, indicated an abnormal record with evidence of severe dysfunction over the left cerebral hemisphere. Subsequent records taken on the 8th, 13th and 20th day following the angiographic reaction (14, 19 and 26 Jan. 1949 respectively) showed considerable improvement, but there was consistent left frontal focal abnormality in the form of 2 to 4/sec. waves with amplitude up to 100 microvolts. On the 25th day following the angiographic reaction (31 Jan. 1949) a record (Fig. 5) was run during a 20-minute period of digital occlusion of the left common carotid without change in the EEG

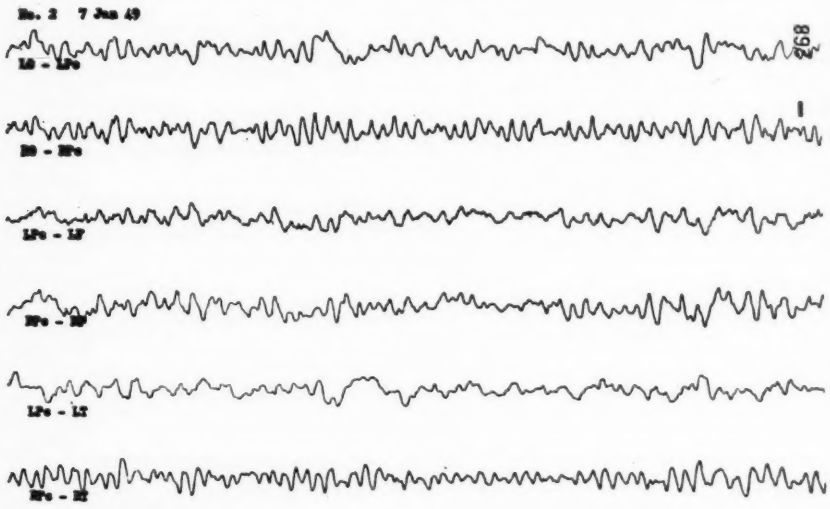

FIG. 4. Record on 1st day after occurrence of right hemiplegia following left cerebral angiography. Slow wave activity is most prominent over left cerebral hemisphere (Case 2).

or EKG, either during occlusion or immediately thereafter. On 1 Feb. 1949 a record run during and immediately after left common carotid ligation failed to show any further difference between the two cerebral hemispheres.

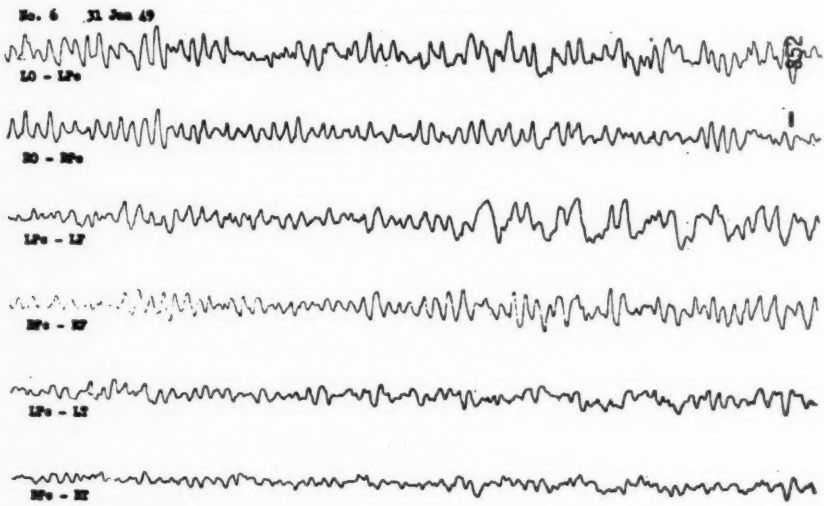

FIG. 5. EEG made on 25th day after onset of transient right hemiplegia showing focal left frontal slowing (Case 2).

PHARMACOLOGICAL ACTION OF AND CLINICAL EXPERIENCE WITH DIODRAST

The chemical name of diodrast is 3,5-diiodo-pyridone-N-acetic acid and diethanolamine. Diodrast is known as well by the trade names of diodone, pyelosin, pylumbrin, perabrodil, and umbradil.[14,16,21] The substance contains 49.8 per cent iodine in firm combination as an organic iodide. It is sterilized by autoclaving and can be resterilized by boiling. According to Marshall[19] the intravenous lethal dose of a 70 per cent solution for the mouse is 5 gm. and for the rat 4.5 gm./kg. of body weight. Further, the tolerated dose for the mouse is 3.75 gm. and for the rat 3.5 gm./kg. of body weight. Doses ranging from the dose tolerated by man to 10 times that amount have been borne well by dogs.[19] Heathcote and Gardner[11] have determined "one equivalent human dose" (1 HD) as 100 mg. of the solid per kg. of body weight. However, these investigators[11] found no toxic effects in dogs following the intravenous administration of 1.5 gm. of perabrodil per kg. of body weight. Perfusion of the isolated rabbit heart produced increased amplitude of contraction with associated increase in coronary flow when solutions of 1:250 strength were used. Similarly, increase in tone of the isolated intestine was observed which was unaffected by atropine. Intravenous doses of 0.75 gm./kg. in the dog caused transient fall in blood pressure and stimulation of respiration. With larger doses, the duration of the blood pressure effect was longer and the initial respiratory stimulation was followed by depression, which, in the fatal cases, was considered the cause of death. In clinical studies, Edwards and Biguria[5] found that the intravenous administration of diodrast, 20 cc. of 35 per cent solution, produced routine fall in blood pressure within 1 minute and reached maximum fall in 5 minutes, with recovery to the original level after 15 minutes. The systolic fall was of the order of 15–20 mm. Hg. and the diastolic fall was 8–10 mm. Hg. They reported one episode of vasospasm with subsequent albuminuria following femoral arteriography with 35 per cent diodrast.

In 1937 Demel and Sgalitzer[2] found evidence to indicate that peripheral arteriography with 20 per cent solution of perabrodil induced prolonged and increased size of the lumen of the finer arterial channels of the limbs of animals. Kristiansen and Cammermeyer[15] could not demonstrate any immediate or delayed histological changes in the vessels or parenchyma of the rabbit brain that could be attributed to 35 per cent perabrodil cerebral angiography. Robb and Steinberg[24] reported studies of angiocardiography with the use of 70 per cent diodrast. The individual dosage employed ranged from 17.5 to 31.5 gm. and 238 intravenous injections were carried out in 127 patients without serious effect. Transient venospasm with pain at the site of injection was noted in a few instances.

In this country diodrast was used initially in 70 per cent concentration for cerebral angiography by Gross,[7,8] but, because of the danger of convulsions, he later recommended its use in the strength of 50 per cent.[9,10] Other

workers[6,13,22,23] have advocated 35 per cent diodrast which is used, perhaps more widely at the present time.

With cinematographic technique, Holm,[13] in 1944, noted smaller calibre of the vessels with 35 per cent perabrodil cerebral angiography than with thorotrast. He interpreted this finding as indicative of increased vascular contraction. Therefore, Holm recommended selection of the contrast medium accordingly, employing thorotrast rather than perabrodil in patients past 50 years of age or when arteriosclerotic and angiospastic conditions were suspected. In 1947 Lindgren[16] reported the use of 35 per cent umbradil (which is identical to perabrodil in composition according to that author) in 153 patients without a major reaction. Green and Arana,[6] in 1948, described their technique of cerebral angiography performed in 107 cases. These workers used a total of 60 cc. of 35 per cent diodrast (21 gm.) in order to obtain the desired vascular phases and projections without major reactions. Diodrast angiography using 35 per cent solution was performed by Ingraham and Cobb[14] in a series of 25 patients with age range of 2 to 67 years without undesirable effects.

Diodrast has been employed extensively in intravenous urography. In 1942 Pendergrass, Chamberlin, Godfrey, and Burdick[21] collected data on 661,800 urographic examinations performed at various centers with different contrast substances. Analysis of questionnaires revealed 26 deaths in addition to those already reported in the literature. There were 10 deaths classified as immediate, occurring within the first 24 hours, all in diodrast injections. In Case II of their report generalized convulsions occurred 8 minutes after intravenous diodrast injection and demise within 45 minutes. Death was attributed to spasm of the cerebral vessels, but no postmortem examination was obtained. Diodrast was not the offending agent in the 16 delayed deaths following urography. A detailed allergic history and use of sensitivity tests were recommended to forestall some of the untoward systemic reactions. Experience with sublingual,[3] ocular,[1] and skin tests[25] for diodrast sensitivity have been reported. Although there is not complete correlation their use prior to diodrast injection appears indicated.

COMMENT

The cases presented followed similar patterns of reaction with complete recovery as determined by clinical neurological examination. In Case 1 no further disturbance of the electrical activity of the brain was present on the EEG taken 6 weeks after the angiographic reaction. In Case 2 there was residual left frontal focal abnormality on EEG 3 weeks later. The clinical manifestations may have been associated with induced vasospasm of the intracranial vessels. Tetraethylammonium chloride has been employed widely both as a diagnostic and therapeutic agent in the relief of certain vasospastic syndromes. This drug was used, therefore, to produce temporary autonomic blockade in the hope of influencing what was thought to be vaso-

spasm, but it is difficult to be certain of the exact role played by tetraethylammonium chloride in these cases.

Although the patients had negative ocular tests for diodrast sensitivity, complications occurred. In addition, these patients underwent subsequent angiographic study without incident. So it would seem that the phenomena were not due to diodrast sensitivity. According to the literature, the quantities of 35 per cent diodrast used, 14 and 17.5 gm. in Cases 1 and 2, respectively, do not seem inordinately large.

It is difficult to obtain information concerning the incidence of transitory hemiplegic phenomena with 35 per cent diodrast cerebral angiography since no reports have been found in a careful review of the literature. Since general anesthesia with endotracheal intubation is used in many clinics to perform cerebral angiography it is possible that the depth of anesthesia depresses vasospastic reflex activity and inhibits the onset of hemiplegia. Further, it is, perhaps, possible that transitory cerebrovascular spasm may occur but its clinical expression be masked during the period of recovery from anesthesia.

SUMMARY

Two cases of hemiplegia following 35 per cent diodrast cerebral angiography are reported. The patients manifested no sensitivity to diodrast by the ocular test. The condition is presumed to be due to cerebral vascular arterial spasm and the patients were treated with tetraethylammonium chloride with recovery. The pharmacological action of diodrast has been considered in the light of its clinical effects.

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ARTERIOVENOUS ANEURYSMS OF THE BRAIN

REPORT OF TEN CASES OF TOTAL REMOVAL OF THE LESION

GÖSTA NORLÉN, M.D.

Neurosurgical Clinic, Serafimerlasarettet, Stockholm, Sweden

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DURING the past 20 years our opinion concerning the treatment of arteriovenous aneurysms has changed radically. Cushing and Bailey,[2] and Dandy[3] considered these lesions practically inoperable and recommended decompression and X-ray treatment. In a monograph on angiomatous malformations and tumours of the brain published in 1936 by Bergstrand, Olivecrona and Tönnis[1] 5 cases are reported in which arteriovenous aneurysms of the brain were successfully completely removed. All the operations were performed by Olivecrona, the first one in 1932. At a meeting of the Scandinavian neurosurgeons in September 1946, Olivecrona and Riives[6] gave an account of experiences gained during the past years (from July 1, 1935) in the surgical treatment of arteriovenous aneurysms of the brain. This report was published in May 1948, and included cases observed at the clinic from July 1, 1935 to August 31, 1946. In 24 of these 43 cases radical excision of the lesion was performed, with 3 fatalities. In the other cases various procedures were carried out, chiefly ligature of accessible arteries and the carotid artery. No appreciable benefit was observed from these procedures and it was felt that ligature of the carotid artery might even be dangerous. Olivecrona and Riives arrived at the conclusion that the choice of treatment lay between removing the lesion and leaving it alone. Recently Trupp and Sachs[10] reported a series of cases of vascular tumours of the brain and spinal cord in which they attacked the lesion with electrocoagulation. By using a very low coagulating current and stroking along the vessel wall, they think it possible to shrink the very largest vessels and then obliterate them. No arteriograms were performed in these cases, but most of them were probably arteriovenous aneurysms. The value of the procedure seems to us a little uncertain. Even if it were possible to coagulate the superficially situated vessels, the main part of the lesion is usually deeply situated and cannot be reached by the coagulating current.

The number of reported cases of the successful removal of an arteriovenous aneurysm is very small. Penfield and Erickson[7] report 2 cases in which a complete block removal had been done. Pilcher[8] reports 3 cases, and Dott[4] 7. From the roentgenograms of Pilcher's cases it appears to us that at least 1 of the cases corresponds to the type of lesion we call a Sturge-Weber's disease, which is a well defined anatomic and clinical entity, and should be differentiated from the arteriovenous aneurysms. Pilcher is of the opinion that most patients with these formidable lesions are amenable to, and should be given, the benefit of radical surgical removal.

The following cases represent all the intracerebral arteriovenous aneurysms removed by the author from July 1946 to December 1948. An attempt at removal of an arteriovenous aneurysm in the left frontal lobe was made in 1939 by the author in a case in which the aneurysm was supposed to be limited to the branches of the middle meningeal artery. The angiographic examination unfortunately had been limited to the external carotid artery and the true nature of the lesion was not recognized until the operation. The patient died on the table from uncontrollable haemorrhage. The case is reported in the paper by Olivecrona and Riives[6] (Case 13).

All the angiograms were performed after percutaneous injection of umbradil (diodrast, perabrodil) into the carotid artery according to the technique described by Wickbom.[11]

Case 1. N. A. N., male, student, aged 19 years. J. Nr. 561/46. In 1942 and 1945 the patient had had attacks of subarachnoid bleeding, the last of which was followed by left-sided hemiplegia. After this there was improvement, but a moderate left-sided spastic hemiparesis, especially of the leg, still remained. He had occasional slight headache. He was admitted to the neurosurgical clinic, July 1, 1946.

Examination. There was a slight left-sided central facial palsy. Moderate left-sided hemiparesis was present, more pronounced in the leg, with slight atrophia. Muscle reflexes were increased and Babinski sign was positive on the left. Optic discs were swollen about 1 D.

Roentgenograms of the skull did not reveal any pathological changes in its structure or vascular channels. Posteriorly, in the right parietal region, about 1 cm. from the mid-line, there was an irregular calcification the size of a pea.

Angiography (Fig. 1A and B). The anterior cerebral artery was enlarged and tortuous, and was the main feeding artery to an arteriovenous aneurysm the size of a walnut, located in the parietal region near the mid-line. From the aneurysm there were some large draining veins to the superior longitudinal sinus. The calcification seen in the roentgenogram was situated in the aneurysm.

Encephalography. There was no dislocation of the ventricular system. The upper part of the right lateral ventricle showed a slight local dilatation, behind which, at the level of the calcification, there was a slight depression of the upper wall of the ventricle. The left lateral ventricle showed no deformity.

Operation (July 16). As a first step, the internal carotid artery in the neck was exposed and a loop put around it, thereby making it possible to strangulate the artery if necessary. After opening the dura, some enlarged arterialized veins were found near the mid-line going to the superior longitudinal sinus. The medial surface of the hemisphere was carefully lifted away from the falx and the aneurysm was removed after ligating some large tortuous feeding arteries from the pericallosal artery. Immediately after extirpation, the patient was able to move his left leg, foot and toes. No blood transfusion was necessary.

Postoperative Course. The patient made a rapid recovery. The day after operation there was a slight weakness and feeling of numbness in the left arm, but these symptoms disappeared after a few days. In the left leg the weakness was perhaps a little more pronounced than before operation. There were no sensory disturbances. The patient left hospital on July 30.

A control examination was done on Sept. 6, 1946. There was a pronounced

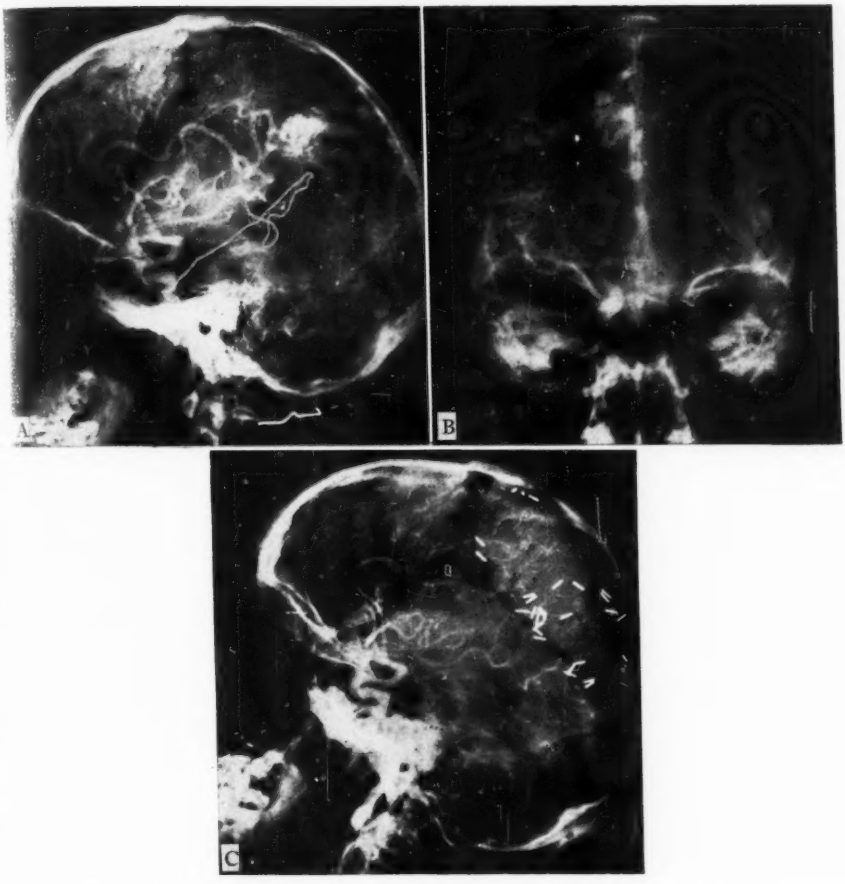

FIG. 1. *Case 1.* Note the difference in size and appearance of the anterior cerebral artery on the angiograms performed 6 weeks after the operation.

improvement of the paresis and the patient was in good condition. Angiography showed that the arteriovenous aneurysm had been totally removed. The anterior cerebral artery was this time of normal size and configuration. The anterior cerebral artery of the left side was also filled with contrast (Fig. 1C).

In November 1948, the patient was still in good condition, and of full working capacity. On one occasion he had had a sudden feeling of numbness with twitchings in the left arm which might have been an epileptic fit. No paresis followed.

Comment. This case was the first in which an angiography was performed after the complete removal of an arteriovenous aneurysm. Some interesting facts regarding the circulation of the brain were observed. The dilated, tor-

tuous, anterior cerebral artery had regained its normal size and configuration. The peripheral parts of the vascular tree were also better filled with contrast than on the first examination. The whole brain circulation had regained its normal appearance after removal of the vascular malformation. The clinical result of the operation must be considered as very good.

Case 2. A. O. H., male, worker, aged 31 years. J. Nr. 291/47. In 1937 the patient suffered a head injury with slight brain concussion. His present illness started in 1945, when, during a bicycle race, he fell and lost consciousness for a short time. This was followed by severe headaches, which continued for 3 weeks, combined with vomiting and impaired vision. He gradually improved. On Feb. 2, 1947, in a ski race, he had another attack of unconsciousness, and was brought to a hospital on the same day. He regained consciousness after some hours, and complained of headache. He vomited, and stiffness of the neck was observed. A lumbar puncture showed increased pressure and haemorrhagic CSF. There was slight papilloedema, but no other neurological signs were found. He gradually improved. On Mar. 24, he was admitted to the neurological clinic, Serafimerlasarettet, and a few days later to the neurosurgical clinic.

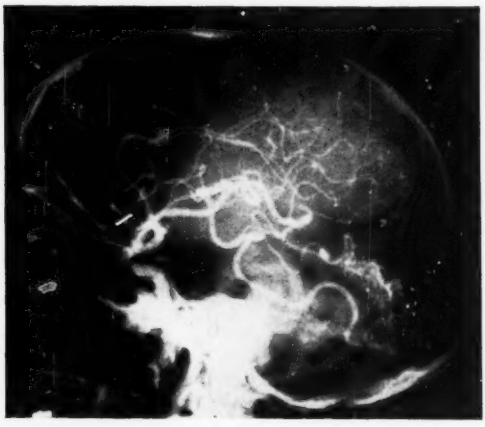

FIG. 2. *Case 2.*

Examination. There was stiffness of the neck. The eye-grounds showed slight choking of the discs. There was left-sided homonymous hemianopia. Slight mental disturbances were noted.

Roentgenograms of the skull were normal.

Angiography (Fig. 2). There was good contrast in the carotid artery and its intracranial branches. In the right parieto-occipital region there was an arteriovenous aneurysm, the size of a plum, with two large feeding arteries coming from the vessels in the Sylvian fissure. From the aneurysm there was a large draining vein to the superior longitudinal sinus.

Operation (April 1). As a preliminary the carotid artery in the neck was exposed and a loop placed round the vessel to permit strangulation during operation if necessary. The aneurysm was located rather close to the under surface of the occipital lobe, about 2-3 cm. lateral of the mid-line. There was also a rather large feeding artery coming from the posterior cerebral artery. The aneurysm reached the posterior horn of the lateral ventricle. This was opened, but there were no vascular connections between the aneurysm and the vessels of the choroid plexus. After the various vessels had been clipped, the aneurysm could be removed. A blood transfusion of 400 cc. was given during the operation.

After practically all big intracranial operations performed under local anaes-

thesia, the patient is left in position on the operating table for 3 hours. The loop around the carotid artery is usually removed immediately after operation, but in this case, because of the patient's prone position, it was to be done after moving the patient from the table to his bed. During this procedure the loop around the carotid artery fastened in something on the operating table, and the artery was torn. Extremely severe bleeding occurred which, after some difficulty, was arrested, and the carotid artery was ligated. The patient's condition was critical for some moments till a blood transfusion had been given. Altogether he was given 8 blood transfusions of 400 cc. each.

The *postoperative course* was then very good. He had no paresis and no sensory disturbances. The left-sided homonymous hemianopia persisted. There were slight mental disturbances of the same degree as before operation. The patient left hospital in good condition on April 19.

Since September 1947 he has had full working capacity in his ordinary work, with improvement in mental symptoms and hemianopia.

Comment. After this complication of a lesion of the carotid artery we abandoned the practice of placing a loop round the carotid artery or applying a vascular clamp during the operation. Further experience and improved technique have shown that this precaution is not necessary. The clinical result of the operation was very good.

Case 3. G. A. B., male, worker, aged 44 years. J. Nr. 331/47. Admitted to neurosurgical clinic, April 11, 1947.

Previous History. Five years previously the patient had complained of double vision, slight pains in the left leg, numbness in the three ulnar fingers of both hands, and weakness of the left arm. During the last years he had sometimes complained of dizziness.

Present Illness. During the night of April 5, 1947, he awakened with severe headache and vomiting. He went to the toilet, and was there found unconscious. He was immediately brought to the neurological clinic.

Examination. The patient was drowsy, but able to answer. Objective findings were: slight left-sided paralysis of the 6th nerve; possibly diminished sensibility for touch on the left side of the face; diminished left corneal reflex; slight left-sided ptosis; pronounced aphasia; homonymous hemianopia to the right; and a right-sided hemiparesis. The discs were not choked. There was no stiffness of the neck; no systolic bruit.

Roentgenograms of the skull were normal.

Angiography. A left-sided arteriovenous aneurysm, the size of a plum, was seen rather deeply situated in the middle of the Sylvian fissure. The feeding arteries were not enlarged, but some dilated draining veins were found (Fig. 3A and B).

On admission the patient was rather drowsy. When he did not improve during the following days, but became more somnolent, it was decided to operate.

Operation (April 16). On opening the dura, the surface of the brain was covered by a subdural haematoma, 1-2 cm. in thickness. There was also an intracerebral haematoma, the size of a hen's egg, in the Sylvian fissure and the temporal lobe. After removing the haematomas with the sucker, some of the feeding arteries could be seen and ligated, and the aneurysm could then be dissected free and removed. Immediately after the patient was able to move his right arm and leg, and also to

answer. One blood transfusion of 400 cc. was given at the end of the operation.

Subsequent Course. The first days after operation the aphasia markedly increased, but then improved gradually. There was no paresis and sensory disturbances were

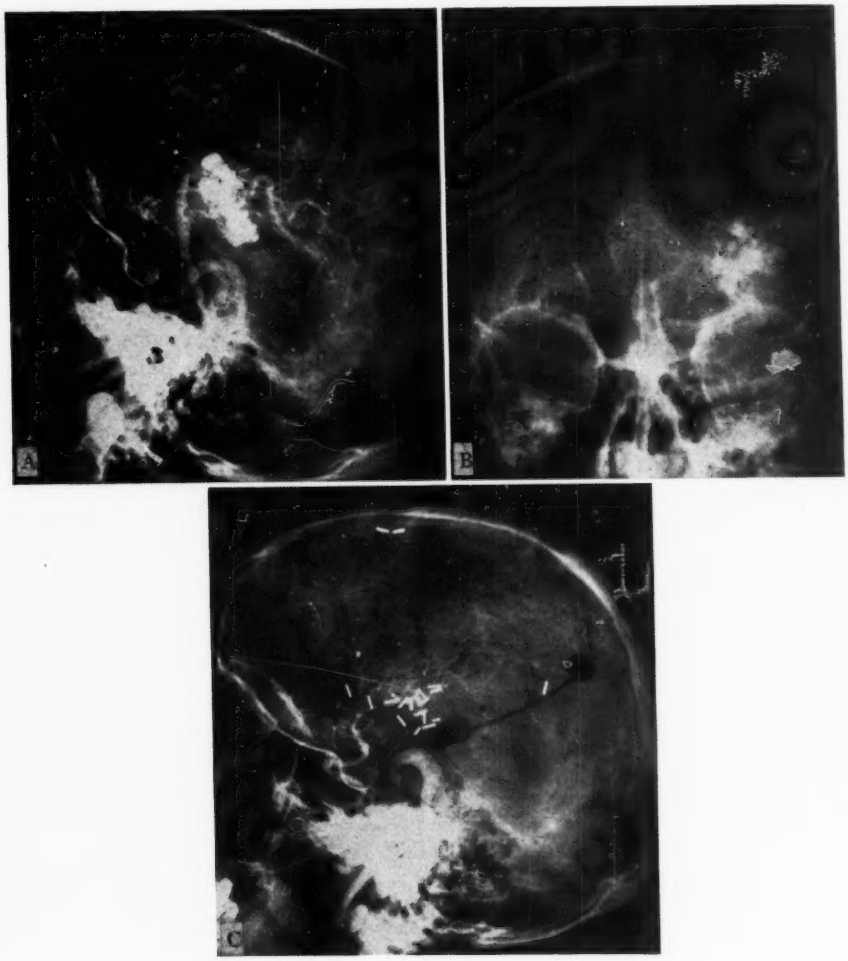

FIG. 3. *Case 3.*

absent. A right-sided pleuropneumonia developed, but he improved and left hospital on May 13, 1947. Angiography showed that the lesion had been totally removed (Fig. 3C). The aphasia had improved, but was still pronounced. In July he had an epileptic fit, and was unconscious for some minutes. The last report was in August 1948. He was still rather aphasic and was not able to work.

Comment. In this case the development of a large subdural haematoma in connection with the rupture of the arteriovenous aneurysm made an urgent operation necessary. An angiography was first done and the lesion considered operable. The clinical end result in this case was not good. The large intracerebral haemorrhage had destroyed too much of the brain tissue. An improvement of the hemiparesis occurred, but the aphasic symptoms persisted and were too pronounced to render the patient capable of work.

Case 4. D. E. A., female, housewife, aged 38 years. J. Nr. 529/47. In 1928, the patient had had head injuries and was unconscious for 24 hours. During the years 1924 to 1930 attacks of headache occurred once or twice a year, rather severe, followed by vomiting. Each attack lasted for 5 to 6 days. Since 1930 these attacks had become more frequent, once or twice a month, and were preceded by something which might be called a visual aura to the right in the visual field. During the last years the headache had been more continuous. Since 1941 there had been weakness and numbness in the left arm and leg. In 1942 epileptic fits occurred in the left side of the body with loss of consciousness.

The patient was admitted to the neurological clinic, Serafimerlasarettet, June 6, 1947.

Examination. Slight weakness of the left arm was found with impaired deep sensibility and astereognosia. There was no aphasia. A slight homonymous hemianopia was evident in the upper part of the visual field to the right.

Roentgenograms of the skull: normal.

Encephalography on air injection after suboccipital puncture: No air got into the ventricular system.

Angiography after injection in the right carotid artery: normal.

The patient was admitted to the neurosurgical clinic on July 7 with suspected brain tumour.

Ventriculography revealed a slight dislocation of the ventricular system to the right. The left temporal horn was dislocated upwards and there was an irregular indentation in the lateral wall of the left lateral ventricle, suggesting an expanding lesion in the posterior part of the left temporal lobe (Fig. 4D).

Angiography in the left internal carotid artery revealed a large arteriovenous aneurysm, the size of a hen's egg, in the left temporal lobe. The circulation of the blood through the aneurysm was very rapid and after a few seconds all the contrast had passed the aneurysm (Fig. 4A and B).

Operation (July 18). Dissection was performed close to the aneurysm, which could be followed practically to the pole of the occipital lobe. Medially the aneurysm reached to the lateral ventricle, and the indentation seen in the ventriculogram corresponded to the large pathological vessels bulging into the lateral wall of the ventricle. There were vascular connections between the choroid plexus and the aneurysm, feeding arteries as well as draining veins. After ligating the vessels, the aneurysm was removed. Five blood transfusions, each of 400 cc., were given during the operation.

Postoperative Course. Immediately after operation the patient was able to move the right arm and leg, and answer questions. The next day she was a little drowsy, with aphasia and right-sided hemiparesis. Subsequently there was marked improvement, and when she left hospital on August 7, there was only a slight sensoric aphasia, right-sided hemiparesis, and right-sided homonymous hemianopia.

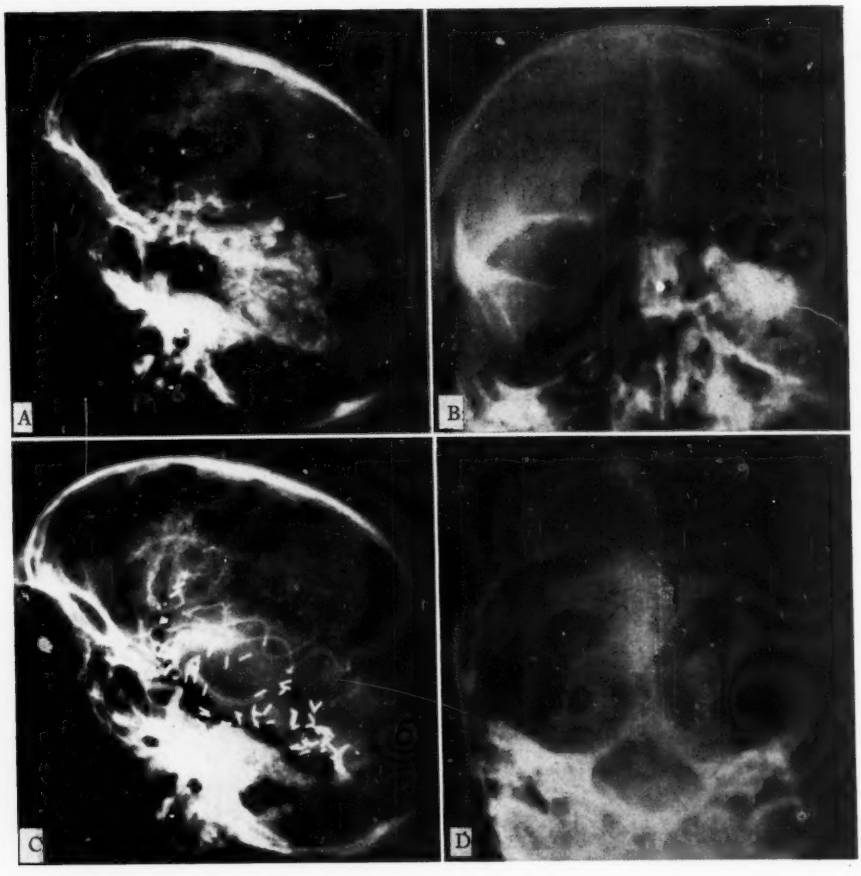

FIG. 4. *Case 4.* Note the difference in contrast filling of the anterior cerebral artery in A and C. In the encephalogram (D) the indentation in the lateral wall of the left lateral ventricle caused by the bulging vessels of the aneurysm is seen.

Arteriography on July 31 showed that the aneurysm had been totally removed (Fig. 4C).

At examination on Aug. 31, 1948, there was marked improvement. Only a slight amnestic aphasia could be observed. There was no right-sided hemiparesis. The left-sided hemiparesis was of the same degree as before operation. There had been some slight attacks of petit mal. She has full working capacity.

Comment. That this formidable lesion in the left temporal lobe was responsible for the Jacksonian fits in the left side of the body is, of course, questionable. If this was the case, and the future might give an answer to this question, it is very remarkable. The clinical result of the operation was very good.

Case 5. B. E. T. S., male, clerk, aged 36 years. J. Nr. 632/47. In 1932 and 1934 he had had head injuries with loss of consciousness.

Present illness started in 1942 with a sudden attack of unconsciousness for 7-8 minutes. Similar attacks occurred 5 to 6 times during the following years. They were unaccompanied by epileptic fits, and there was no paresis afterwards. Since 1946, 2 to 3 times a month he had had twitching in the left side of the face, with a feeling of numbness and paraesthesia, also noted in the left arm. For the last months headache of varying degree had been present, and during the past year, slight mental disturbances. He was admitted to the neurological clinic, Serafimerlasarettet, Mar. 31, 1947.

Examination. Neurological findings, aside from partial 3rd nerve paralysis on the left side, were negative. On auscultation over the right maxillary sinus a short bruit was heard synchronous with the pulse.

Roentgenograms of the skull and *encephalography:* normal.

Angiography. There was good contrast in the Sylvian vessels, but none in the anterior cerebral artery. In the posterior part of the right frontal lobe, about 1 cm. above the Sylvian fissure, an arteriovenous aneurysm, the size of a plum, was seen with some dilated feeding arteries from the Sylvian vessels, and a very enlarged draining vein going to the superior longitudinal sinus. In the middle of the temporal lobe there was another smaller arteriovenous aneurysm (Fig. 5A and B).

As the symptoms were very slight it was decided not to operate and he was discharged from hospital. However, as the epileptic fits increased in frequency, the patient, after a few months, became very anxious about his condition and wanted to take the risk of operation.

Operation (Aug. 28, 1947). The brain surface was very vascular with enlarged tortuous arteries. One of the arteries was followed down into a brain sulcus, where the aneurysm could be grasped with forceps and lifted out after the feeding arteries and the draining vein had been ligated with silver clips. The same procedure was then carried out with the other aneurysm. There were several large vessels between the two aneurysms. Some of these were extirpated but others had to be left alone. These vessels were very fragile and it was difficult to stop the bleeding from them.

Postoperative Course. During the first few days there was a paresis of the left hand with numbness. This, however, rapidly improved, and when the patient left hospital on September 17, there was only a very slight weakness and numbness left, more especially in the thumb.

Angiography, September 15, showed that both aneurysms had been removed but that between them there were still some widened pathological vessels (Fig. 5C).

The patient started to work in January 1948. On control examination in September 1948, he still had some headache, but not to the same degree as before operation. He had had 3 attacks of epileptic fits of Jacksonian type localized to the left leg and arm. He was a little restless and nervous.

Angiography, Sept. 27, 1948, showed that the widened, pathological vessels between the two aneurysms had disappeared (Fig. 5D).

Comment. An unusual case in which two aneurysms were removed at one stage. An arteriogram 1 year after operation revealed the interesting fact that the dilated pathological vessels between the two lesions, observed immediately after the operation, had disappeared.

Case 6. A. W., female, housewife, aged 30 years. J. Nr. 108/48. In 1946 and 1947

the patient had had attacks of subarachnoidal haemorrhage with complete recovery. On Jan. 8, 1948, another attack occurred. She was admitted to the neurological clinic.

Examination. The patient complained of severe headache. There was no bruit.

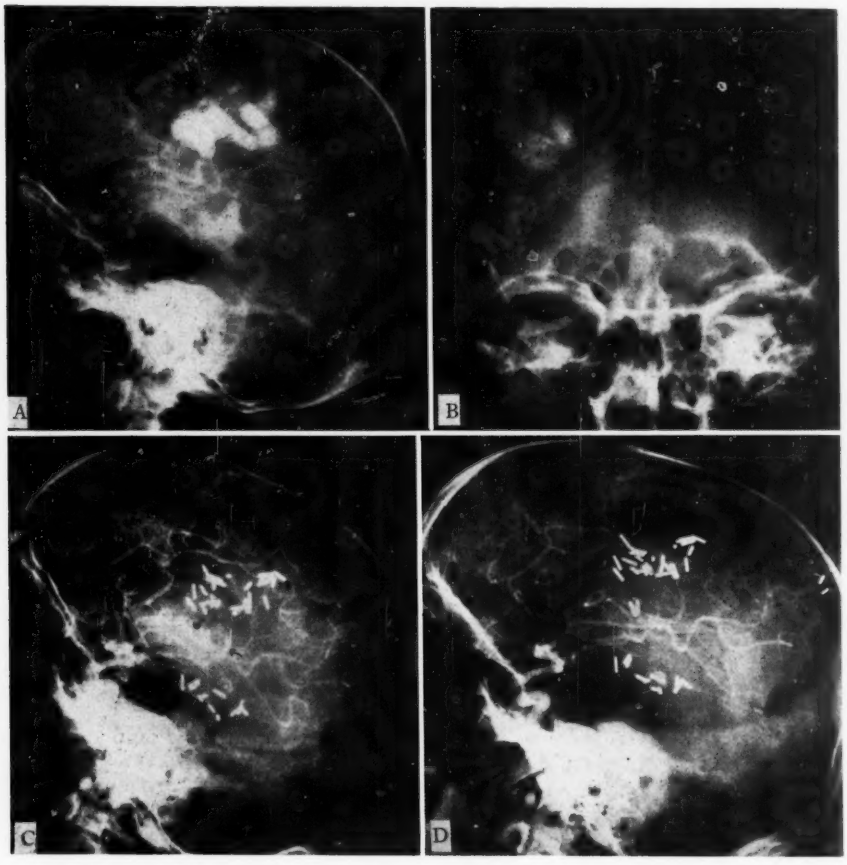

FIG. 5. *Case 5.* The contrast filling of the anterior cerebral artery, which is practically lacking in the first examination (A) is clearly seen on the angiograms 2 weeks after operation (C). In the latter some pathological vessels between the aneurysms are still evident, but on an angiogram 1 year after the operation (D) these vessels have a normal appearance.

Neurological findings were: pronounced stiffness in the neck; and homonymous hemianopia to the left. After some days the hemianopia disappeared.

Angiography revealed an arteriovenous aneurysm, the size of a mandarine, in the right parietal lobe. Most of the feeding arteries came from the Sylvian vessels. There were also some from the anterior cerebral artery. There were widened draining veins to the superior longitudinal sinus. The aneurysm reached rather deeply into

the brain tissue, and the deepest point was only 2 cm. from the mid-line (Fig. 6A and B).

The patient was admitted to the neurosurgical clinic on Feb. 2, 1948, where an operation was performed the same day.

Operation. One of the feeding arteries was followed into the depth of the brain tissue where the aneurysm was found surrounded by sclerotic discoloured brain

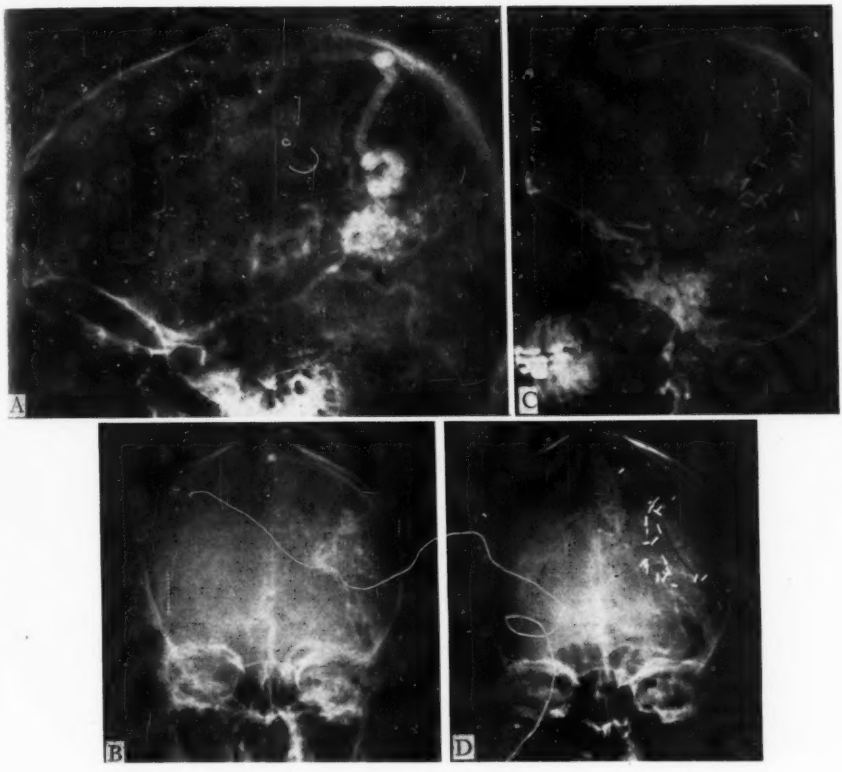

FIG. 6. *Case 6.*

tissue indicating that haemorrhages had occurred. The dissection of the aneurysm was therefore rather difficult. After ligating the feeding arteries around the aneurysm and the draining vein, the aneurysm could be removed.

Postoperative Course. The patient made a rapid recovery. There was no postoperative paresis, and she was able to leave hospital on February 17. The day before, an *angiography* had been performed which showed that the arteriovenous aneurysm had been totally removed. The feeding arteries were now of normal size (Fig. 6C and D).

On control examination on May 21, the patient was in a very good condition,

and of full working capacity. There were no subjective symptoms, no paresis, and visual field was normal. The clinical result of the operation was excellent.

Case 7. E. A. A., male, farm-labourer, aged 34. J. Nr. 257/48. In 1924 the patient had a head injury. Since 1925 he had had epileptic fits, with numbness in the right hand and arm, and right side of neck, combined with loss of consciousness for 5 minutes. There was no paresis afterwards. Slight mental symptoms were noted. He had no headache.

Examination. Neurological findings were normal.

Roentgenograms of skull and *encephalography:* normal.

Angiography. In the left parietal lobe an arteriovenous aneurysm, the size of a walnut, was found. Particularly one of the feeding arteries from the Sylvian vessels was enlarged and tortuous. One widened draining vein to the superior longitudinal sinus was seen. There was no contrast in the anterior cerebral artery (Fig. 7 A and B).

Operation (April 1). The large feeding artery was ligated close to the aneurysm which could then be dissected free from the surrounding brain tissue and removed after ligation of the draining vein.

Postoperative Course. The day following the operation there was no paresis, but this gradually developed in the right side, together with aphasia, and the patient had repeated Jacksonian fits. These symptoms disappeared, and when the patient left the hospital on April 24, there was only a slight weakness in the right arm, more pronounced weakness in the right foot, and slight hyperaesthesia of the right foot and leg. Muscle reflexes on the right side were increased. Babinski sign was negative bilaterally.

Angiography on April 17 showed that the aneurysm had been totally removed. The dilatation of the feeding artery had diminished (Fig. 7 C).

On examination in October 1948, the hemiparesis had completely disappeared. The patient still had epileptic fits of the same type as before operation, but more frequent.

Angiography on October 13, showed that the former dilated feeding artery was now of normal size. The anterior cerebral artery was now clearly seen and of normal size. Before operation it had not filled with contrast, and 3 weeks after operation was still very thin (Fig. 7 D).

Comment. The hemiparesis and aphasia that appeared some days after operation were probably due to a local oedema. The angiography done half a year after operation showed that the brain circulation had regained its normal appearance. From a clinical point of view the frequency of the epileptic fits had increased. In spite of that we are of the opinion that a lesion of this kind, with this localization, even if the clinical symptoms are very slight, should be removed. A rupture of this aneurysm might have given an intracerebral haemorrhage with destruction of brain tissue causing an irreparable hemiplegia. The patient is of full working capacity.

Case 8. K. S. H., aged 40, factory hand. J. Nr. 293/48. Since 1932 this patient had had Jacksonian fits in the right side of the body, starting in the right foot. The frequency of the fits had increased during the last years. The first years he lost consciousness in connection with the fits, but not during the latter years. After the attacks

he had a feeling of numbness in the right leg. Following an attack on Mar. 27, 1948, there was paresis in the right arm and leg, and the next day he became somnolent. He was admitted to the neurosurgical clinic on April 9.

Examination. Neurological findings were: pronounced right-sided hemiparesis

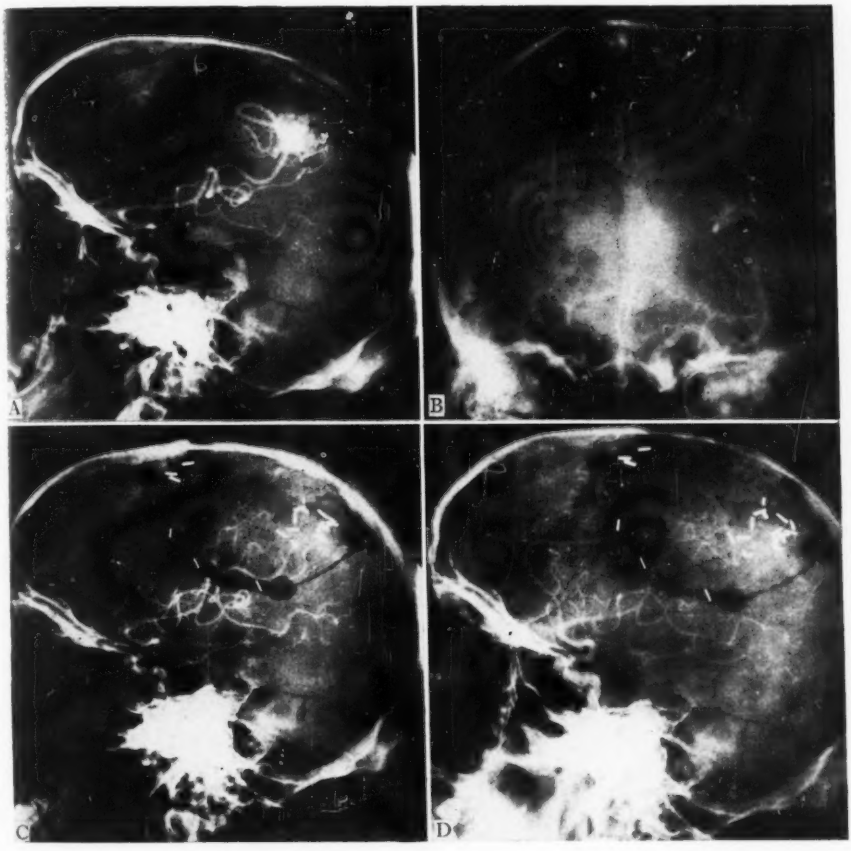

FIG. 7. *Case 7.* The main feeding artery, 3 weeks after the operation, is still dilated (C), but on an angiogram (D) 6 months later the artery has regained its normal size and appearance.

with total paralysis of the arm; and bilateral choked discs. There was no aphasia.

Roentgenogram of the skull: normal.

Angiography after injection into the left internal carotid artery: in the parietal lobe an arteriovenous aneurysm the size of a mandarine was found. The lesion was localized near the mid-line, and received its arterial supply both from the anterior and middle cerebral arteries. There were draining veins to the superior longitudinal sinus (Fig. 8 A).

An angiography was also performed after injection into the right internal carotid artery. There was contrast in both anterior cerebral arteries, and the aneurysm was clearly seen also in this examination (Fig. 8 B).

Encephalography after suboccipital puncture revealed a slight dilatation of both lateral ventricles, a little more pronounced on the right side. The upper wall of the

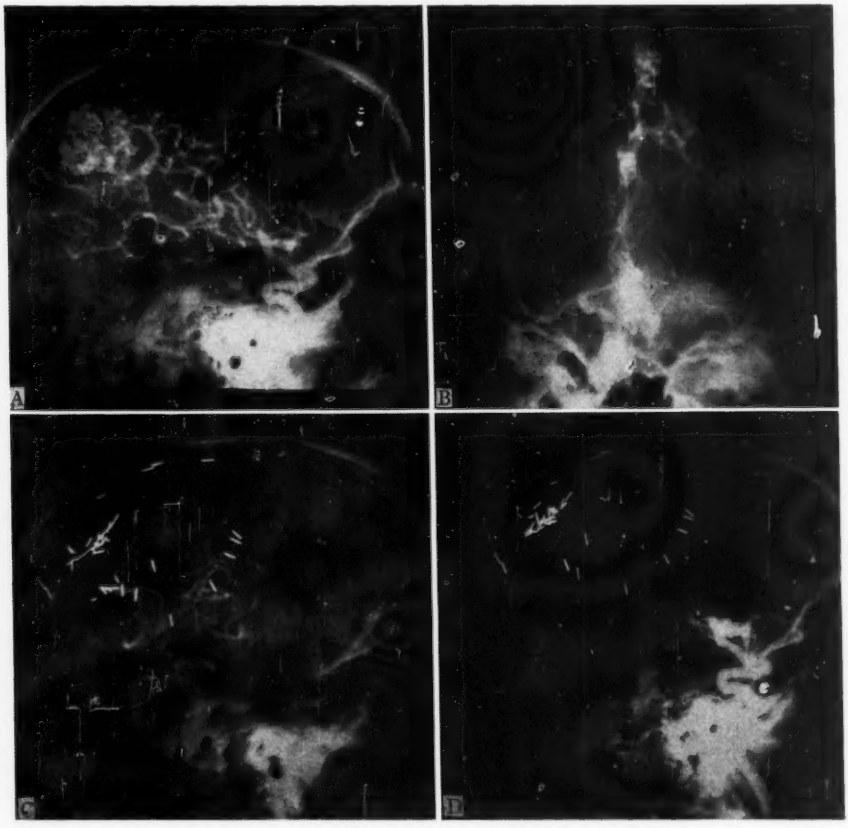

FIG. 8. *Case 8.* The arteriovenous aneurysm in the left hemisphere is filled with contrast (A) and is also seen after injection in the right carotid artery (B). On an angiogram 3 weeks after the operation there are still some dilated vessels (C), and the anterior cerebral artery is enlarged, but 9 months later the pathological vessels have disappeared and the anterior cerebral artery is of normal size (D).

left lateral ventricle was a little depressed, corresponding to the localization of the aneurysm.

Operation (April 22). On the medial surface of the hemisphere some feeding arteries from the anterior cerebral artery were ligated. The dissection of the aneurysm was rather difficult, and was complicated by a lesion of one of the enlarged

draining veins. After removal of the aneurysm there was some bleeding from dilated vessels surrounding the aneurysm. The only possible method of checking that bleeding was to put a piece of spongostan on the bleeding surface and leave it in place. On turning down the flap a lesion of the superior longitudinal sinus had occurred, so it was necessary to suture it and cover the sinus with some pieces of spongostan. Eleven blood transfusions, each of 400 cc., were given during the operation.

Postoperative Course. The first days there was a right-sided hemiplegia but in a few days the movements of the leg returned. There was no aphasia.

Angiography on May 14 showed that the aneurysm had been removed. Dilatation of the anterior cerebral artery persisted partially. The enlargement of the feeding arteries from the middle cerebral artery had diminished. The patient left hospital on May 21 (Fig. 8 C).

On examination on August 24, a pronounced improvement was seen. He was able to walk around with a stick and move the right arm slightly. He had had two small epileptic fits with twitchings in the right leg without loss of consciousness. In February 1949 he was able to walk around without a stick but the right arm was still rather paretic. *Angiography* showed that the pathological vessels had disappeared and the anterior cerebral artery had regained its normal size (Fig. 8 D).

Comment. A postoperative angiographic study of the brain circulation shows that vessels of abnormal size and configuration regain their normal appearance after the removal of an arteriovenous aneurysm.

Case 9. V. M. S., aged 52, female, housewife. J. Nr. 544/48. For 1½ years before admission the patient had observed that the left part of the visual field sometimes disappeared for a few seconds. In the summer of 1947 she had an attack of rather severe headache to the right in the occipital region, with stiffness in the neck. After a week she was all right again. There were no other symptoms. On Mar. 16, 1948, she had a new attack of headache, slight dizziness, and subjective left-sided homonymous hemianopia. After a few days the headache increased, combined with vomiting and stiffness in the neck. She was admitted to the neurological clinic, where a left-sided homonymous hemianopia was observed, and a lumbar puncture revealed a sanguinolent CSF.

Roentgenogram of the skull showed, on both sides, rather large vascular markings, but no other changes in its structure and no destruction. The foramen spinosum was rather large but of the same size on both sides.

Angiography was performed on several occasions in the carotid arteries on both sides. On the right side, close to the mid-line in the occipital region, there was a small aneurysm, and a little more laterally a vascular saccular malformation the size of a pea was seen. From this region there were some dilated draining veins. The feeding arteries to this vascular malformation appeared to be two enlarged tortuous branches from the external carotid artery, probably the posterior branch of the middle meningeal artery, one on each side (Fig. 9 A and B).

The lesion was considered to be an arteriovenous aneurysm in the right occipital lobe.

Encephalography. There was no dislocation or deformation of the ventricular system. In the right occipital lobe near the mid-line, corresponding to the localization of the arteriovenous aneurysm, there were signs of local atrophy.

Operation (July 19). The middle meningeal artery was dilated. No abnormal vascular connections between the dura and the brain were seen. An incision was made 2–3 cm. laterally from the mid-line where the brain tissue was a little softer than usual. A cystic formation, the size of a walnut, was found communicating with

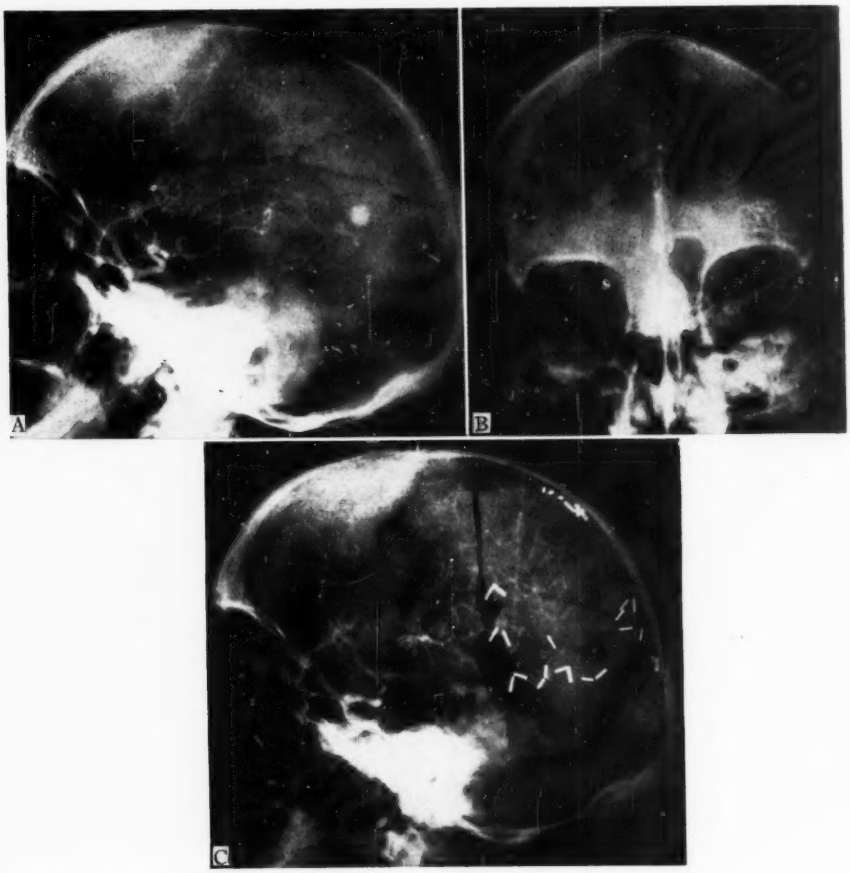

FIG. 9. *Case 9.* Of the whole aneurysm only a small saccular malformation is filled with contrast. A thrombotization of the rest of the aneurysm had occurred.

the ventricular system. In the medial wall of that cyst there was a brownish-green, rather dense formation, the size of a walnut, which looked like an organized haematoma. From the middle of this formation there was a very large vein going to the superior longitudinal sinus. No enlarged feeding arteries could be seen. It was very easy to remove the whole formation.

Microscopical examination revealed an arteriovenous aneurysm with large

vessels of arterial type with very thick walls surrounded by an organized haematoma.

Postoperative Course. The patient made a good recovery. No paresis but a left-sided hemianopia was still present when the patient left hospital on August 5.

Angiography on August 2 showed that the vascular malformation had been removed (Fig. 9 C).

Comment. The angiographic study in this case was rather difficult, and it was not so easy to come to a conclusion regarding the localization of this lesion. A thrombotization and organization of the aneurysm had occurred. The clinical result was excellent and the patient is of full working capacity.

Case 10. E. K. P., female, aged 29, housewife. J. Nr. 930/48. In 1940 a sudden attack of right-sided hemiplegia occurred. The patient was admitted to hospital, where a lumbar puncture revealed a subarachnoidal haemorrhage. There was a marked improvement during the next months and after 6 months she was all right again. Since 1943 she had had attacks of paraesthesias in the right arm, and since March 1944, epileptic fits of Jacksonian type, starting in the right arm, with loss of consciousness. These attacks had occurred about 3 times a year, but the last year they had become more frequent.

She was admitted to the neurosurgical clinic on Nov. 10, 1948.

Examination. The eyegrounds were normal. Slight weakness of the right arm and disturbance of the deep sensibility of the right foot were found. Muscle reflexes were increased and Babinski sign was positive on the right side.

Roentgenograms of the skull: normal.

Encephalography revealed a slight dilatation of the left lateral ventricle.

Angiography. Above the posterior part of the Sylvian fissure an arteriovenous aneurysm, the size of a mandarine, was seen. The lesion reached down rather deep, the deepest part being only $\frac{1}{2}$ cm. from the mid-line. There were enlarged feeding arteries, mainly from the Sylvian vessels, but also some feeding vessels from the anterior cerebral artery. Widened draining veins went to the superior longitudinal sinus (Fig. 10 A and B).

Operation (November 17). Some dilated vessels were seen on the surface but the aneurysm was rather deeply situated close to the wall of the lateral ventricle, with vascular connections with the choroid plexus. Posteriorly the aneurysm was limited by a cavity which had been formed by the previous haemorrhage. Because of the deep localization of the aneurysm and the vascular connections with the choroid plexus the removal of the lesion was rather difficult. At the end of the operation there was a pronounced motoric aphasia and complete hemiplegia.

Postoperative Course. During the days following operation there was a gradual improvement of the hemiplegia, and when the patient left hospital on December 10, she was able to walk around but there was still a paresis of the hand and fingers. The motoric aphasia persisted for a week but then gradually improved. On discharge from hospital no aphasic disturbances could be observed.

Angiography on December 7 showed that the aneurysm had been removed. The size of the feeding arteries was now normal (Fig. 10 C and D).

Comment. The size and localization of this lesion made an attempt at removal rather hazardous. On the other hand, another rupture of the aneurysm might have caused irreparable hemiplegia and aphasia. The patient

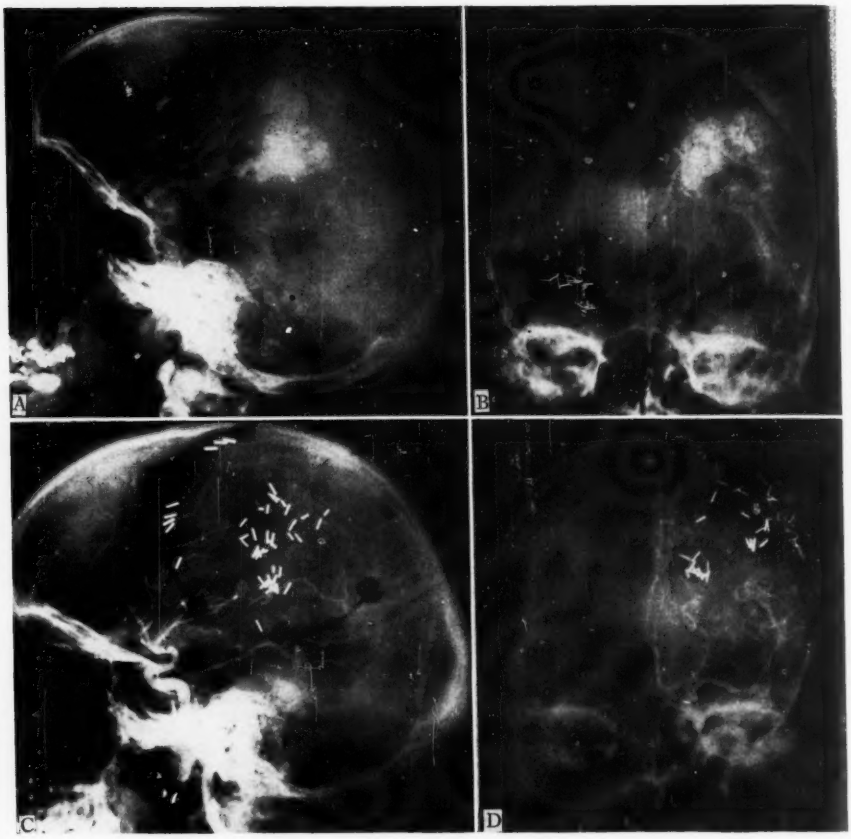

FIG. 10. *Case 10.* In the frontal picture (B) the connection between the anterior choroidal artery and the aneurysm is clearly seen.

was informed of the situation but wanted to take the risk of an operation. The ultimate result cannot yet be judged but the postoperative course speaks in favour of a good result.

DISCUSSION

In considering the indications for operation, Olivecrona and Riives have pointed out that in the end probably most, if not all, patients with these lesions die of haemorrhage or are completely incapacitated, and that the mental deterioration which is a common feature in cases of an inveterate lesion also strengthens the argument in favour of removal of the lesion. During the last years the indications for operation have been widened, and rather large aneurysms, even in the left hemisphere, have been successfully

removed (Cases 5 and 10). The poor prognosis for epileptic cases with an inveterate lesion speaks in favour of an early operation in those cases where epilepsy is the only symptom. In Case 7 there was a fairly long history of epilepsy as practically the only symptom. In this case removal was considered easy because a fairly small aneurysm was localized superficially. Even if the prognosis for epilepsy was poor, the operation was considered justified. It must be borne in mind, however, that indications for operation must be judged with the greatest criticism. Pilcher recommends radical surgical removal of these lesions but adds that, nevertheless, the surgeon must be able to recognize the limits of such possibilities and must have the courage to withdraw when faced with lesions beyond these limitations.

The clinical result of the operation has been very good in 7 patients and all are of full working capacity (Cases 1, 2, 4, 5, 6, 7 and 9). This is anticipated also in Case 10. In 2 cases (Cases 3 and 8) an improvement has occurred when the symptoms before and after operation are compared, but both are invalids, one with pronounced aphasia and the other with hemiparesis. Even if a further improvement is possible, the destruction of brain tissue from the intracerebral haemorrhage has been too large to permit the patient to regain full working capacity.

The most interesting thing in this series was the angiographic study of the brain circulation and the appearance of the brain vessels before and after removal of the lesion. In most of the cases there was a very rapid return to the normal, usually within 2–3 weeks, of vessels which before the operation were enlarged and tortuous. In other cases pathological vessels were still visible in the angiogram after that time, but a few months later they regained their normal appearance (Cases 4, 7 and 8). From angiograms performed before operation it was evident that the contrast filling of vessels not belonging to the group of vessels that went to the aneurysm was very poor. This might indicate poor nutrition and anoxemia of these parts of the brain, which might cause brain atrophia and be responsible for the mental symptoms which are common in inveterate cases. After removal of the aneurysm, the contrast filling of the vessels of these parts of the brain was good and clearly seen (Cases 1, 4 and 5).

Reid[9] has pointed to the fact that even very large angiomas, either spontaneous or as a result of inflammation, thrombosis or slight surgical intervention, may disappear. He reports a case of an extensive angioma involving the entire leg, which was cured, by thrombosis, after the excision of a very small portion of it. In Case 9 a thombotization of practically the whole aneurysm had occurred, indicating that this aneurysm might have been spontaneously cured. That angiography should precede surgical intervention in cases of aneurysms has been emphasized by many authors (Tönnis,[1] Leriche[5]) and is quite clear.

Angiography following operation gives valuable information not only about the completeness of the removal but also about the brain circulation and the condition of the brain vessels.

SUMMARY

1. Ten cases of total removal of an arteriovenous aneurysm are presented.

2. In 9 cases angiograms have been performed after the removal.

3. The postoperative angiographic study shows that the brain circulation regains its normal appearance and the brain vessels return to normal size and configuration after removal of an arteriovenous aneurysm.

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CLINICAL ANALYSIS OF EIGHTY-EIGHT CASES OF METASTATIC CARCINOMA INVOLVING THE CENTRAL NERVOUS SYSTEM

WITH AN OUTLINE OF THERAPEUTIC PRINCIPLES

ARTHUR R. ELVIDGE, M.D., AND MAITLAND BALDWIN, M.D.

Department of Neurology and Neurosurgery, McGill University and the Montreal Neurological Institute, Montreal, Canada

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A CLINICAL analysis of 88 consecutive neurosurgical cases of metastatic tumour involving the central nervous system is presented. The primary and secondary sites of growth are compared with reference to the relative frequency of the anatomical location. The pathological types are noted, and the general methods of treatment are outlined.

PRIMARY SITES OF GROWTH

The primary sites of growth listed in order of frequency of appearance in the series are given below:

Lung and Bronchus	20
Breast	14
Kidney	4
Thyroid	3
Rectum	3
Prostate	3
Spine (multiple myeloma)	2
Ovary	1
Bladder	1
Uterus	1
Colon	1
Cervix	1
Adrenal	1
Nasal Mucosa	1
Parotid	1
Pancreas	1
Testicle	1
Doubtful	29
Total	88

As noted above, no accurate definition of the primary location was made in the last 29 cases. However, the lesions in the central nervous system were carcinomata. Failure to accomplish localization of the primary was due to incomplete clinical studies, or incomplete autopsy examination.

PRIMARY PATHOLOGICAL TYPES

In the lung and bronchus there were 8 adenocarcinomata, 5 cases of spindle cell carcinoma and 4 squamous cell carcinomata. Three cases were not analyzed as to cell type.

In the breast there were 7 adenocarcinomata, 2 carcinoma solidum, 3 scirrhous carcinoma, and 2 undiagnosed by cellular type.

In the thyroid there were 2 cases of adenocarcinoma, and 1 case unspecified.

Those of the G-U tract are listed as follows:

Uterus—Adenocarcinoma.
Cervix—Squamous epithelial carcinoma.
Ovary—Carcinoma simplex.
Testicle—Teratoma.
Kidney—3 hypernephroma, 1 alveolar adenocarcinoma.
Prostate—3 adenocarcinoma.
Urinary bladder—1 transitional cell carcinoma.

Those of the G-I tract showed the following cell types:

Pancreas—Adenocarcinoma.
Colon—Adenocarcinoma.
Rectum—3 adenocarcinoma.

The parotid gland showed 1 adenocarcinoma. The adrenal cell type was not specified. There was 1 mucous cell carcinoma arising from the nasal mucosa.

The predominant histopathological type in this series is adenocarcinoma.

SECONDARY SITES OF GROWTH

Tumours arising from the lung and bronchus metastasized to the following parts of the central nervous system, in order of frequency: to the cerebrum, 11; to the cerebellum, 5; to the vertebral column, 1. Two cases showed metastasis to both cerebrum and cerebellum, and 1 case showed metastases to the skull and spine.

There were 12 tumours arising in the right lung or bronchi. These showed metastases to both the right and left sides of the brain and to the vertebral column, as follows: right cerebrum, 5; right cerebellum, 1; left cerebrum, 1; left cerebellum, 2; vertebral column, 2. One tumour in the right upper lobe showed metastases to the right cerebellar hemisphere at operation and to the left cerebrum at autopsy.

Six tumours arising from the left lung or bronchi, showed secondary foci as follows: metastases to the right cerebrum, 1; to the right cerebellum 1; to the left cerebrum, 3. One tumour from the left upper lobe showed metastases to both the right cerebrum and right cerebellum at operation. One from the left lower bronchus showed metastases to both sides of the skull, and to the bodies of T7 and 8 and the pedicles of T10 and 12.

From origins in the breast, the following metastases occurred: to the cerebrum, 9; to the cerebellum, 1; to the meninges, 1; to the pons, 1; to the pituitary, 1; generalized to the cerebrum and longitudinal sinus, 1. From the right breast these neoplasms spread as follows: to the left cerebrum, 2; to the right cerebrum, 1; to the left cerebellum, 1; to the left temporal meninges, 1; to the pons, 1; to the pituitary, 1. The left breast showed metastases with the following locations: to the right cerebrum, 3; to the left cerebrum, 2; generalized, 1. One case of adenocarcinoma in the right breast of a male metastasized to the pituitary.

There were 3 cases of metastases from the thyroid, all to the thoracic spine (T3, T3 and T6–9 respectively).

From the genito-urinary system secondary lesions were noted as follows: from the bladder (1 case) to the spine (T6–9); from the prostate (3 cases) to the cervical and to the thoracic spine respectively; from the cervix uteri to the lumbar and sacral vertebrae, at L3–4–5 and S1. (An adenocarcinoma of the uterus showed no clearly defined metastatic lesion, but cerebral thromboses were noted.) From the ovary (1 case) to the spine (T3); from the left testicle to the left temporal and left cerebellar areas. The peri-renal tumours metastasized as follows: from both kidneys (1 case) to the thoracic spine (approximately T6); from the right kidney (2 cases) both to the left cerebellum; from the right kidney (1 case) to the left frontal area. The adrenal tumour metastasized to the 2nd and 3rd lumbar vertebrae.

In the gastro-intestinal group, the pancreatic lesion metastasized to the 1st and 2nd lumbar spine. The carcinoma of the rectum showed secondary lesions in the right cerebellar hemisphere, with extension to the right middle fossa and the right frontal meninges. The carcinoma of the colon also metastasized to the right cerebellar hemisphere.

The tumour of the left parotid metastasized to the left middle fossa. The mucosal carcinoma of the nose developed a left frontal cerebral lesion.

There were 2 cases of multiple myeloma. One arising from the 5th thoracic vertebra showed metastases to the skull, the ribs and the right femur, and the other, whose primary lesions were discovered in the 2nd and 3rd cervical vertebrae, showed multiple metastatic lesions in all bones.

Summary of Secondary Sites of Growth. (1) In the cases of carcinoma of the lung or bronchus and breast, there was no apparent correlation in the above information, of side of primary focus to side of metastasis. Thus, it cannot be stated (at least from observation of these cases) that a tumour arising in one or the other lung field would metastasize to one or the other cerebral hemisphere. However, from a clinical point of view the predominance of metastatic spread from the lung field was to the cerebral hemisphere as opposed to the cerebellum or spine. This was true of both bronchogenic carcinoma and carcinoma of the breast.

(2) Primary carcinoma arising from the thyroid, bladder, prostate, cervix, ovary, adrenal gland, kidney (1 case with bilateral involvement) and pancreas metastasized to the spinal column. In 1 case of primary carcinoma

of the right kidney, metastasis occurred to the left frontal lobe and in the 2 other instances to the left cerebellar hemisphere. From the left testicle spread occurred to the left temporal lobe and the left cerebellum. One tumour of the colon and 1 of the rectum metastasized to the right cerebellar hemisphere. Growth from the nasal mucosa on the left side metastasized to the left frontal lobe, and from the left parotid lesion to the left middle fossa.

TABLE 1

Age and sex incidence

	Male	Female	Age
Lung	17	3	47.3
Breast	1	13	52.9
Thyroid	0	3	27
Bladder	1		68
Uterus		1	56
Ovary		1	50
Colon		1	56
Spine	2		50.5
Testicle	1		47
Kidney	4		46.5
Prostate	3		71
Cervix		1	58
Adrenal	1		58
Parotid	1		62
Pancreas	1		41
Rectum	2	1	50.5
Nasal Mucosa		1	39
	34	25	51.8

In this series (Table 1) the ratio of males to females was 3 to 2. This excludes a group of 29 cases in which the primary lesion was not definitely identified as to location or type. The average age was 51.8 years. In tumours of the bronchus or lung 17 males were affected as compared with 3 females, whereas in tumours of the breast 1 male was affected as compared with 13 females. All the tumours of the thyroid were in females and all the tumours of the urinary tract were in males. The ratio of males to females in tumours of the genito-urinary tract was 3 to 1, and the tumour of the adrenal was in a male.

FAMILY HISTORY OF CARCINOMA

Information available in the cases under discussion is not adequate to allow a conclusion on the incidence of carcinoma in patients' relatives. However, there is evidence of carcinoma in the families of a few widely scattered cases.

DOUBTFUL PRIMARY FOCI

Twenty-nine cases in the series are considered separately, because information as to the exact location of the primary tumour is not available. This deficiency exists because adequate clinical data were lacking or because in

certain cases in which the patients died after a brief hospital course, autopsies were not granted. The cases are briefly summarized for their general interest.

There are 8 cases of spinal metastases. Examination of the surgical specimens in these cases did not suggest the primary focus except in 1 instance of metastasis to the 9th thoracic vertebra, which resembled a hypernephroma. One patient with metastasis to the body of the 6th thoracic vertebra, is known to have had a mass in the left breast, which was not biopsied in this hospital. One with metastasis to the lower cervical spine was re-admitted a year later with ascites. No biopsy was obtained, but a presumptive clinical and radiographical diagnosis of carcinoma of the large bowel was obtained. Fifteen showed metastases to the cerebral hemisphere and examination of the surgical specimen in each of these showed that a large proportion were epithelial in type and some were adenocarcinomata. One tumour which metastasized to the right occipital lobe resembled a carcinoma of the bowel. Three cases showed secondary lesions in the cerebellar hemisphere. Examination of the pathological specimen in one of these suggested ovarian carcinoma, and in another, adenocarcinoma resembling bronchogenic origin.

REMARKS ON THE TREATMENT OF METASTATIC CARCINOMA IN NEUROLOGICAL SURGERY

Metastatic carcinoma as a therapeutic problem in neurological surgery presents the necessity of the alleviation of increased intracranial pressure, the relief of pain and the prevention of paraplegia.

Cerebral Metastasis. In cases where the intracranial metastasis is single, removal should be considered for the relief of the intense headache apart from any improvement of neurological signs. Pneumonectomy for primary carcinoma of the lung by Dr. C. A. McIntosh resulted in a patient returning to work for 2 years. A secondary growth was then removed from the right temporal lobe. Patient left hospital in 2 weeks, ambulatory and well.

On the Treatment of Pain. The treatment of pain is possible through the employment of definitive surgical techniques. In the majority of patients cordotomy will relieve pain from the trunk and lower limbs, and is the method of choice if there is no compression of the spinal cord or nerve roots at the vertebral level. When the procedure is carried out with care and accuracy, there need be no disturbance of bladder function. The operation may be done unilaterally with an interval before carrying out section of the other side. Though generally considered to be more hazardous there are cases where it may be desirable to do both sides at the same operation, and the authors believe that it can be done bilaterally in one procedure without disturbance or alteration of bladder function or sphincter control. At the discretion of the operator the section can be made in the dorsal or upper cervical region. The selection, of course, is dependent largely upon the level of the pain source, taking into consideration afferents passing by way of the sympathetic system. After laminectomy and exposure of the cord, the dentate

ligament is identified, gently picked up and a fine knife inserted 2 mm. anterior to it and parallel to the fiber tracts in the desired direction through the anterolateral segment of the cord. It is turned at right angles and drawn out laterally.

In cases where spinal exploration and decompression are undertaken it has been the custom of the authors to make extradural spinal root sections at the level of the growth both to relieve and prevent pain. The number of roots selected for sectioning, usually 2 on each side, is judged by the amount of involvement and the proximity of growth to the roots. In all cases explored and decompressed by the authors in the last few years extradural root section has been employed and the pain, which is frequently bandlike, was relieved at once. In these cases cordotomy is generally not necessary.

In cases requiring a very high level of sensory anesthesia for relief of pain, high cervical cordotomy may be employed but it seems too difficult to be certain of obtaining a satisfactorily high level, for example, above the 6th dermatome. The writers have employed a combined procedure in this rare type of case, illustrated as follows:

Case 1. Posterior root section of the 2nd to the 6th cervical roots inclusive on the ipsilateral side and high cordotomy at the C2–3 level on the opposite side was performed. It was possible in this case to bring the sensory level of the cordotomy up to meet the loss in the hand from the root section. The junction occurred at the 3rd digit. It was interesting to note the difference in type of sensory loss in the two areas. Pain and temperature sense was lost over all spinal sensory dermatomes on the right side from C2 and below, while over the index finger and thumb slight loss of 2-point discrimination was added. A 3 mm. spread of a caliper was appreciated normally over the volar surface of the distal phalanges of the 3rd, 4th and 5th digits as compared to a gap of 4 mm. over the index finger and thumb. There was no loss of power except that already present due to rather massive oedema from lymphatic obstruction and though there was some difficulty in manoeuvering the arm, sense of position was grossly intact. Sensation to touch was preserved over the hand approximately to the elbow. Above the elbow on the lateral side of the arm (C6 and up) touch was not felt. Section of the upper 6 (2–6) posterior roots did not appear to disturb function greatly probably because the small muscles of the hand derive innervation from roots C8 and D1 and the afferents no doubt correspond.

It is not the purpose to discuss the question of methods of treatment of pain in the region supplied by the 5th and the 9th cranial nerves and upper cervical supply. Most cases seen by the writers have involved principally the trigeminal distribution. For these, alcoholic injection of the maxillary and mandibular branches seemed to be reasonably satisfactory and simple. Otherwise one must resort to root sections by any of the usual methods. Intramedullary trigeminal tractotomy has been advocated by a few.

CONCERNING THE TREATMENT OF PARALYSIS

When paralysis occurs or is imminent, spinal decompression by laminectomy and partial removal of the neoplasm is indicated to prevent progres-

sion. A paraplegic patient with metastases will pose a difficult nursing problem because, in addition to the complex care which the patient with carcinoma or primary malignant disease of the spine requires the treatment necessitated by disturbance of function of the urinary bladder and anal sphincter would be included. Occasionally with decompression and removal of tumour disappearance of paraplegia will occur. It should be stressed that laminectomy is not only of temporary benefit but in some instances may appreciably increase the life expectancy of the patient as well as contribute materially to his comfort during that time. In illustration, the case of a patient who is listed in the above series is cited.

Case 2. Five separate operative procedures were carried out on this woman. The 1st of these, which included intradural posterior root section (D7, 8, 9, right) gave complete relief of paralysis and pain for 8 months. The 2nd operation (mostly blood cyst, very little tumour found), after 8 months, also relieved the paralysis completely and gave her an almost equivalent pain-free interval. The 3rd, 12 months later, which included extradural root section (D7, 8, left), and the 4th operation, after another 9 months, again cleared up the paralysis and relieved the pain. On the 5th admission, 3 months later, for recurrence of the growth partial removal was repeated also with further root section. The patient succumbed at the end of this procedure.

This patient experienced more than 2½ years of reasonably comfortable life. She was provided with a brace when walking. Without laminectomy in the first instance, followed by the other procedures, her illness would have been complicated by paraplegia, much more pain and the related problems of bladder and sphincter disturbance.

SUMMARY

1. Eighty-five clinical cases of carcinoma with metastasis to the central nervous system are reviewed, with some suggestions as to treatment.

2. The majority of patients with carcinoma of the bronchus (lung) or breast sought neurosurgical consultation or treatment as a result of metastases to the cerebral hemispheres.

3. The tumours of the thyroid, bladder, prostate, cervix, adrenal, pancreas and kidney (1 case) exhibited clinical symptoms and signs from metastases to the spinal column.

4. The average age incidence for the series was 51.8 years.

5. The ratio of males to females was 3 to 2 in the general series. In the tumours of the lung, 16 males were affected as compared with 3 females; while in tumours of the breast, 1 male was affected as compared with 13 females.

6. Neurosurgical treatment should be directed toward the relief of: (a) intracranial pressure; (b) pain; (c) paraplegia.

7. The removal of a solitary intracerebral metastasis in selected cases may be attended with satisfactory relief of symptoms.

8. The authors have found it desirable and wish to point out the value

of simultaneous extradural section of spinal roots which are, and which are apt to be, compressed in cases justifying a decompressive laminectomy for impending paraplegia with partial removal of neoplasm. This procedure generally obviates the necessity for cordotomy.

9. In the special instance of diffuse pain of the upper extremity that does not respond to roentgen therapy, a posterior root section (C2–C6) combined with cervical cordotomy has given satisfactory relief.

10. It seems desirable to relieve spinal compression by decompressive laminectomy to prevent or minimize paralysis. Five such operations were performed in 1 patient over a period of 3 years with satisfactory result.

THE SURGERY OF THE CRANIOPHARYNGIOMAS*

PHILIP D. GORDY, M.D., MAX M. PEET, M.D., AND
EDGAR A. KAHN, M.D.
Department of Neurosurgery, University Hospital, Ann Arbor, Michigan

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THERE continues to be justifiable pessimism among neurosurgeons as to the surgery of the craniopharyngiomas. This has been occasioned by the high operative mortality and the large number of cases in which total removal is manifestly impossible. It is the purpose of this paper to analyze the results in 51 consecutive cases of patients operated upon for craniopharyngioma at the University Hospital following the publication of Peet[7] in 1927 from the same source.

The operation at this particular institution has invariably been approached with the object in mind of total extirpation. This was believed to have been accomplished in 25 of the 51 cases. The fact that 6 of the 17 who survived what was thought to have been total excision have died of recurrence is disconcerting. It means that one cannot be as certain of total removal in this inaccessible type of brain tumor as one can be with others such as the psammomatous meningiomata.

One thing, however, is certain. Where firm adhesions have not formed between the tumor and adjacent vital structures, such as the optic nerves, the carotid arteries and the floor of the third ventricle, complete removal is possible and is sometimes surprisingly simple. This means then, that if the results of the surgery of the craniopharyngiomas are to be improved the diagnosis must be made earlier.

It is not the purpose of this presentation to discuss again the embryology, pathology and all of the clinical manifestations of this particular tumor. Rather, we should like to present various aspects of these cases in relation to their surgical end results.

AGE INCIDENCE

The youngest patient in this series was 2½ years, the oldest 65. The largest number of patients were in the first and second decades (Table 1).

SEX INCIDENCE

No specific sex incidence was demonstrated, there being 27 males and 24 females.

CLINICAL MANIFESTATIONS

The manifestations of increased intracranial pressure were a predominant part of the picture presented by the younger patients, especially those in group I. Headaches, vomiting and papilledema were seen frequently, either

* Presented at meeting of the Harvey Cushing Society in San Francisco, California, August 21, 1948.

separately or in association with each other. In addition, there were other evidences of disturbed or altered function such as listlessness, drowsiness, irritability and generalized weakness. With increase in age and the approach of puberty, localized neurological disturbances, and defects in growth and metabolism and sexual maturation became a more prominent part of the picture than increase in intracranial pressure. Decrease in visual acuity and field defects occurred in a high percentage of cases after the first decade. It

TABLE 1

Age group with fundus findings

Group	Age (Years)	Number	Fundus
I	1–10	17	3 normal 5 primary optic atrophy 8 papilledema 1 secondary optic atrophy
IIa	11–15	8	1 normal 4 primary optic atrophy 1 secondary optic atrophy 1 Foster-Kennedy 1 papilledema
IIb	16–20	4	1 normal 2 primary optic atrophy 1 papilledema
III	21–30	5	3 papilledema 2 secondary optic atrophy
IV	31–40	8	4 normal 3 papilledema 1 secondary optic atrophy
V	41–50	6	5 normal 1 primary optic atrophy
VI	51–60	2	1 normal 1 primary optic atrophy
VII	61–70	1	normal

has been pointed out by previous authors[2] that the symptomatology in the first decade tends to be predominantly that of increased intracranial pressure. Often, of course, the first thing that is noted is that the child appears to have difficulty in seeing. The child may also be small for his age. With increasing age, localized pressure on the optic nerves results in demonstrable field defects.

Of the 17 patients 10 years or under, only 6 were believed by the examiner to be normal in regard to bodily habitus. Seven patients were variously

described as being either "slightly obese" or exhibiting the Fröhlich's syndrome. Four were small and underdeveloped. One female child of 8 showed a rapid onset of sexual development with the presence of pubic and axillary hair.

In the patients in group II, with the onset of puberty, disturbances of growth and endocrine imbalance appeared more prominent. Only 1 of the 12 was thought by the examiner to be normal in growth, development and habitus. The remaining patients fell into two general groups: those tending towards the Fröhlich habitus and the second characterized by underdevelopment and the appearance of pituitary dwarfism. Four fell within the first of these two groups and 5 in the second.

The patients in the groups from 21–55 showed a high incidence of endocrine disturbances with amenorrhea, loss of libido and poorly developed secondary sex characteristics. The patients in the last two age groups showed no growth or endocrine disturbances.

Diabetes insipidus was a frequent finding both preoperatively and postoperatively. Twelve patients had this in the preoperative period; of the 12, 6 were 10 years of age or under, and 3 were 20 years of age or under. Twenty patients had polyuria and polydipsia following operation. In some instances this was transient, but in the majority of the cases it was a more long-standing difficulty. One patient, living today 8 years postoperatively, continues to have a moderate degree of diabetes insipidus.

There was, of course, in all age groups a high degree of ocular and visual abnormalities. Diplopia was a frequent finding with either a unilateral or bilateral 6th nerve paresis. This represented another non-localizing sign of increase in intracranial pressure. Abnormalities of the fundus (Table 1) and the visual fields were present in a high proportion of cases. The field defects ranged from complete blindness to homonymous and heteronymous defects. The bitemporal and homonymous types of field defects were found without predominance of one or the other.

Neurological deficits are not usually considered to be a predominant part of the clinical signs that these patients present. Love[5] states that the neurological examination in his series of 11 cases was usually negative. However, of this group of 51 cases, 29 showed some degree of positive neurological findings. These included palsies of the 5th, 6th and 7th cranial nerves, nystagmus, hemiparesis, impaired coordination and Babinski responses.

X-RAY FINDINGS

X-ray examination of the skull is of great aid in supplementing the clinical examination of the patient. A large number of these tumors are sufficiently calcified so that the diagnosis can be made with certainty from the plain film of the skull. In the group of 35 cases reported by McKenzie and Sosman,[6] 71 per cent of the roentgenograms showed calcification, and 76 per cent showed an abnormal sellar outline. In our series of 51 cases, 65 per cent showed suprasellar calcification or suprasellar calcification combined with

intrasellar calcification and in 63 per cent of the cases the sella showed some degree of abnormality.

The changes seen, exclusive of intrasellar and suprasellar calcification, were intrasellar and extrasellar erosion, depression of the floor, thinning of the anterior and posterior clinoids, erosion of the dorsum sellae and ballooning of the sella turcica. In 8 cases, or 15 per cent, the plain film of the skull was negative. Air studies were done on 20 patients. These included pneumoencephalogram and ventriculogram. In 1 case (JA 39) the skull x-ray and a pneumoencephalogram were negative. The diagnosis in this case rested on the clinical findings of bitemporal hemianopsia, headache, blurred vision, nausea and vomiting, amenorrhea and drowsiness.

ELECTROENCEPHALOGRAPHIC FINDINGS

In 4 of the more recent cases the EEG* was called into play as a diagnostic aid.[1] One case (AS 48) was of especial interest in this regard since the entire history consisted of grand mal and petit mal epilepsy which had been present 24 years and 10 years respectively. A localizing EEG on a 3-channel machine in this instance showed minor generalized epileptic features. The background pattern was of a strong alpha type with abortive spike-and-wave and fast components. The localizing waves were of irregular 2 to 3 / sec. high voltage type, spikey or superimposed by alpha waves coming from the right temporal region. From all types of recording, including common mid-parietal bipolar and contralateral ear ground hook-up, the preoperative interpretation was that there was a lesion in the right hemisphere either in the right temporal lobe or anterior and adjacent to it with some extension superiorly and posteriorly. At operation a tumor was found below and anterior to the chiasm and extending back to the hypothalamus. A later examination of the preoperative EEG record revealed suppression of deltas bipolarly in anteroposterior right midtemporozygomatic arch recording and right parietolateral-premotor recording. It also revealed disturbance of the potential pattern of the lateral aspect of the left anterior regions when the potentials of those regions were led across the tumor area to the right ear alone. These considerations would place the lesion in the medial aspect of the anterior temporal lobe and inferiorly and would not suggest its extension superiorly and posteriorly, as was thought preoperatively.

A second semi-localizing EEG study (MD 46) showed evidence (1 to 2 / sec. waves) of a lesion in the left hemisphere. This was left midtemporal, deep and was thought probably to be a neoplasm. Many 6 to 7 serial waves were found in the background. Vertex common bipolar lead and, particularly, alternate right and left ear ground leads gave some idea of the depth and medial placement of the lesion. The tumor was approached on the left through the lateral ventricle. Partial removal of a large tumor was accom-

* Electroencephalograms were interpreted by Dr. B. K. Bagchi of the Electroencephalographic Laboratory of the Neuropsychiatric Institute and University of Michigan Hospital.

plished. The patient died in the immediate postoperative period and autopsy showed no tumor in the 3rd ventricle but some residual tumor in the hypothalamus.

In the third case (AB 12), a routine EEG work-up on a 3-channel machine showed very high voltage 2 to 2½ and 3/sec. serial waves mainly in both prefrontal regions, particularly right prefrontal, and both occipital regions and very little in the motor and temporal regions, against a background of 5 to 6/sec. waves. The occipital waves were interpreted as a delayed effect from deep anterior regions (?chiasmatic). The over-all evaluation was abnormal with evidence of a deep lesion in the midline spreading in many directions. At operation a cystic suprasellar lesion was found.

A localizing EEG examination on an 8-channel machine in the fourth case (DH 19) showed minor but clear bilateral abnormality (medium voltage fast waves anteriorly) but no definite localization. Using nasal and right ear ground vectors, rare but very definite slow undulations were seen in some anterior leads which were referable to deep midline region anteriorly, but preoperatively sufficient emphasis was not placed on them and hence they were regarded as not being consistent with or specific enough for an adequate localization. A cystic lesion beneath the chiasm was found at operation. This was aspirated at the first procedure. At a second procedure 40 days later the tumor was removed except for a solid portion below and posterior to the chiasm.

These examinations were done in only a few instances and not as part of the routine work-up. There are, however, indications that a complete EEG study may be of definite value in those cases with obscure clinical findings and negative plain films of the skull.

SURGICAL TREATMENT

The surgical treatment of craniopharyngiomas is accompanied by a high operative mortality rate in most of the published reports. Love, Shelden and Kernohan[5] reported a series of 11 consecutive cases in which operation was successfully performed. There has been, however, no adequate follow-up report as yet on these patients, and in most of the cases no attempt at radical removal was made.

The factors contributing to the high mortality of this particular type of lesion are, of course, related to the position of the growth in reference to the hypothalamus and carotid arteries. Often the tumor is of large size when the patient is first seen by the neurosurgeon, rendering complete removal difficult and hazardous.

Certain authors[3] have felt that the high mortality rate does not justify an attempt at complete removal of the tumor but insist, rather, that evacuation of the cyst should be done and irradiation then carried out to decrease the activity of the secretory cells. Ingraham and Scott[4] recently reported on a group of 16 patients with craniopharyngioma treated at The Children's Hospital and the Peter Bent Brigham Hospital between 1932 and 1945. They

advocate partial removal of the wall of the tumor with evacuation of the contents of the cyst. This, they state, can be carried out in most instances without mortality. Six patients had shown no evidence of recurrence for 1 year or longer while 1 had shown no progression of symptoms for 13 years. They stated that "In general, it seems logical to conclude that the patients with late recurrence, who have had a relatively long remission following partial removal of the cyst, should be subjected to a repetition of surgical treatment, whereas those with early recurrences and without contraindica-

FIG. 1. Photographs showing location of incision (Case 2).

tion such as serious reduction of visual acuity, might best be treated by withholding surgery and instituting roentgen therapy."

It has been the policy at the University Hospital to approach the surgery of the craniopharyngiomas with the thought in mind of performing a complete excision whenever this was permitted by the local situation. The operative approach that has been followed here is a right frontotemporal osteoplastic flap (Fig. 1). Depending on the operating surgeon, the anterior limb is brought to the midline, just at the hairline, or is placed somewhat more laterally in a line with the pupil. The cases were about equally divided between those approached intradurally, and those approached extradurally by opening the dura along the lesser wing of the sphenoid. Love, Shelden and Kernohan[5] have felt that the tumor could be most satisfactorily approached when the flap was turned on the side corresponding to the eye that showed the most marked visual loss. They advocated operation on the right when visual loss was equal.

We invariably approach the tumor from the non-dominant side of the brain, even though this may be the side of best preserved vision. It is actually easier to avoid injury to the optic nerve when it is exposed prior to attackin

the tumor than it is to preserve intact a nerve hidden behind the tumor mass. It is always easier to protect a vital structure from injury when once it is exposed and visualized.

In this series of 51 cases, complete excision was thought to have been accomplished by the operator in 25 instances. In 5 more cases the completeness of excision was questionable. In 21 cases the surgeon had to content himself with less than a complete removal. The amount of tumor tissue left in these instances varied from a small shred of capsule left adherent to one of the carotid arteries to a large mass of solid tumor lying behind the posterior clinoid processes or pressing up into the 3rd ventricle.

The operative mortality in this group of cases was 41 per cent. The mortality in most of these instances could be traced to severe hypothalamic reaction attendant upon operative trauma. Eleven patients died later from recurrence at intervals from 3 months to 16 years (Table 2). The evidence for recurrence was based on either postmortem findings, re-operation or a follow-up history showing death to be the result of an intracranial lesion.

TABLE 2

Patients dying of recurrence (37 per cent of 30 patients surviving operation)

Patient	Age	Estimated Removal	Survival Period	Pathology
O.G.	14	Complete	16 yr.	Adamantinoma
B.C.	5	Partial	11 yr. 4 mo.	Cystic adamantinoma
R.J.	3	Complete	10 yr.	Cystic adamantinoma
F.G.	4	Complete	9 yr. 6 mo.	Cystic adamantinoma
W.K.	8	Partial	5 yr.	Cystic adamantinoma
D.U.	29	Complete	2 yr. 9 mo.	Cystic adamantinoma
E.H.	50	Partial	2 yr. 8 mo.	Cystic adamantinoma
J.W.	15	Complete	1 yr. 4 mo.	Cystic adamantinoma
S.W.	3	Partial	4 mo.	Adamantinoma
B.B.	31	Partial	3 mo.	Cystic adamantinoma
J.M.	4	Complete	13 mo.	Adamantinoma

Thirty per cent of those patients in whom complete removal was thought to have been accomplished died in the immediate postoperative period. Of those with partial removal 52 per cent died in this period. This discrepancy, which is quite at variance with other published figures, may be explained.

Partial removal did not, in this group of cases, mean aspiration of the cyst with removal of a small portion of its wall. The patients tabulated as having had a partial removal were those in whom complete extirpation was attempted but could not be carried out because of the large size and inaccessibility of the tumor as well as adherence to vital structures.

Twelve of the 51 patients, or 23.5 per cent, are known to be alive at the present time. The follow-up on these cases varies from 10 months to 10 years (Table 3).

It may be seen that total extirpation, successfully carried through, is

attended by a significant proportion of long survivals. The best results are to be expected in the younger patients. The cases in the first two decades are those in which the tumor is most likely to be small, cystic and enucleable. As the age of the patient increases a larger proportion of the tumors encountered are large, solid tumors with extension posterior to the sella and into the 3rd ventricle.

Four case summaries will be presented. Three of these cases illustrate the favorable results that may be obtained in the younger patients with

TABLE 3

Patients now living (23.5 per cent of 51 cases)

Patient	Age	Removal	Survival Period	Pathology	Present Condition
G.H.	3	Complete	10 yr.	Adamantinoma	Excellent
G.G.	19	Complete	7 yr. 7 mo.	Cystic adamantinoma	Fröhlich syndrome
J.A.	39	Complete	6 yr. 3 mo.	Adamantinoma	Excellent
K.W.	19	Complete	6 yr.	Rathke's pouch cyst	Good. Diabetes insipidus
R.C.	12	Complete	5 yr. 6 mo.	Adamantinoma	Good. Vision improving
Y.L.	17	Partial	4 yr.	Squamous epithelioma	Good. No menses
M.M.	9	Complete	2 yr. 9 mo.	Craniopharyngioma	Good
D.P.	3	Partial	2 yr. 6 mo.	Adamantinoma	Good
		Complete	1 yr.	Adamantinoma	Good
C.M.	52	Complete	2 yr. 3 mo.	Adamantinoma	Excellent
R.C.	31	Complete	2 yr. 3 mo.	Craniopharyngioma	Excellent
B.T.	3	Complete	10 mo.	Adamantinoma	Good. Diabetes insipidus
E.L.	2½	Complete	10 mo.	Adamantinoma	Excellent. Regaining vision

small, enucleable lesions. Case 4 is a follow-up report on one of Peet's original cases.

CASE SUMMARIES

Case 1. B.T., a 3-year-old negro girl, was admitted on Sept. 4, 1947 because of polyuria, polydipsia and poor appetite. In January 1947, 9 months before admission, her mother first noted that the right eye crossed at times, the eye turning outward. At these times the child would shake her head and blink as though she were having difficulty in focusing. At the same time she began to have polyuria to such an extent that her bed was frequently wet. Marked polydipsia was also present. In March, 3 months after the initial onset of symptoms, the child had several episodes of vomiting and complained of a frontal headache. The diagnosis of craniopharyngioma was made in another hospital. Laboratory studies showed a dilute urine with a specific gravity of 1003. This rose to 1010 on the progressive concentration test. BUN, urea clearance, blood calcium and phosphorus and serum proteins were normal. Glucose tolerance test showed a fasting level of 35 mg. per cent, 132 at ½ hour, 171 at 1 hour, 154 at 2 hours and at 3 hours the level had fallen to 58. Skull films showed an enlarged sella with a shell of calcium within the sella. The chest film was normal. Past and developmental histories were entirely normal. She was sent to the University Hospital for surgery.

Examination. She was a slender, underdeveloped negro child who appeared

quite bright and interested in her surroundings. Height and weight were below the normal for her age. The right superior rectus was overactive. It was not possible to do visual fields and the optic fundi were normal. Review of the outside x-rays in this hospital showed that the enlargement of the sella turcica was of the "balloon" type. There was a thin shell of calcification within the sella (Fig. 2). This extended superiorly above the upper extremity of the dorsum into the suprasellar region. Roentgenograms of the long bone ossification centers were normal. A diagnosis of craniopharyngioma was made.

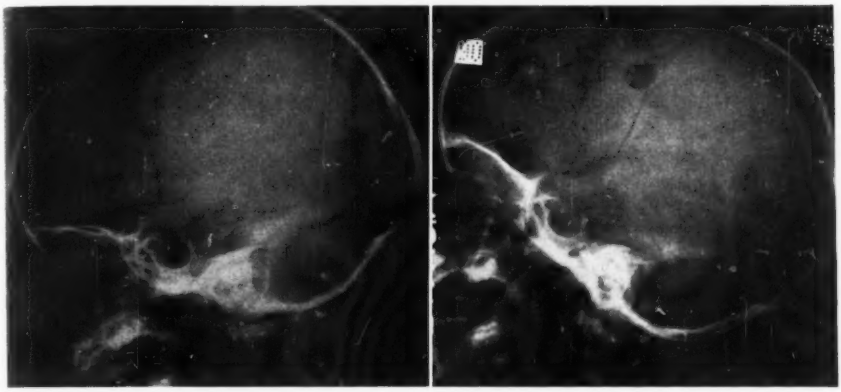

FIG. 2. Case 1. *Left*, Pre-operative x-ray showing intrasellar calcification. *Right*, 9 months postoperative, showing complete removal.

Operation. On Sept. 8, 1947 a right frontotemporal osteoplastic flap was turned. The dura was definitely under increased pressure. This was relieved by tapping the ventricle and the dura was then opened along the lesser wing of the sphenoid. The frontal lobe was retracted and the right optic nerve was then seen with a bluish, cystic tumor bulging medial to the nerve. The cyst was opened and contained characteristic yellow fluid. It was about the size of a hickory nut. In piecemeal fashion, it was entirely removed from the region of the left optic nerve and chiasm, which had been markedly compressed, and from the right optic nerve, although it was somewhat adherent to the internal carotid artery. It was thought that perhaps a small amount might have been left at this point. When the tumor was removed the pituitary gland could be seen in the depth of the sella. Closure was effected in the usual manner.

Course. Immediately postoperatively, there was hyperthermia to 103°, which was controlled. The patient alternated between marked restlessness and marked lethargy. Convalescence proceeded well except for several episodes of left-sided Jacksonian convulsions. On Sept. 12, 1947, the operative site was reexplored and 15 to 20 cc. of blood were removed from the extradural space. There was none around the chiasmal region. By Sept. 14 the convulsions had ceased and the patient was beginning to take fluids well. On Sept. 18 she was up and about. She was placed on pitressin tannate, $\frac{1}{4}$ cc. in oil, which controlled the diabetes insipidus well, and was discharged on Nov. 5, 1947, alert and cheerful.

On Dec. 15, 1947 she was examined in the Out-Patient Department. Her height was $37\frac{1}{2}$ inches, her weight $25\frac{3}{4}$ pounds, and she was doing well. She was seen again 7 months later and was in approximately the same condition. She has remained on her medication.

Microscopical Diagnosis. Adamantinoma.

This case illustrates the favorable result that may be expected from the removal of a small, cystic lesion that can be peeled away from the carotid arteries and optic nerves without undue trauma to the hypothalamus. Complete removal would very definitely seem to be the procedure of choice in this type of tumor.

Case 2. G.H., a 3-year-old white female child, was admitted to the University Hospital on June 27, 1938. Three months previously she had fallen and struck the right frontal region. For the last 4 months she had complained of pain in this area, showed loss of interest, and appeared to be more or less distracted. There was no history of vomiting, visual impairment or disturbance in growth.

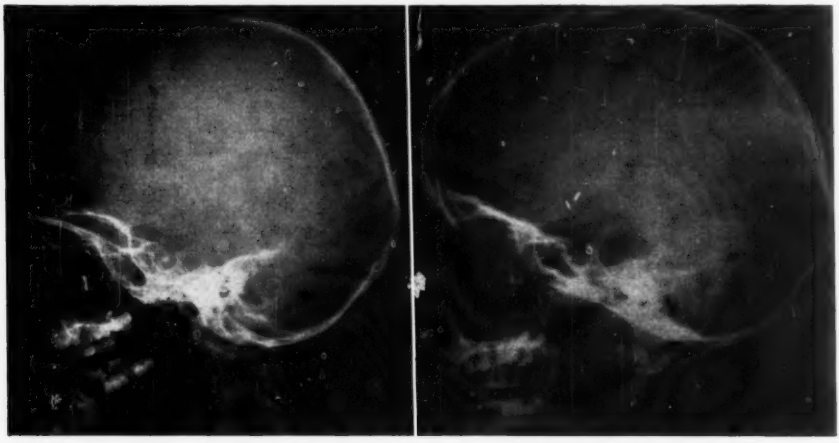

FIG. 3. Case 2. *Left,* Pre-operative x-ray showing intrasellar calcification. *Right,* 9 years postoperative, showing complete removal.

Examination. The patient was a well-developed child, not appearing acutely ill. Neurological findings were negative except for a bilaterally positive Babinski.

Ophthalmological findings were normal; it was not possible to obtain visual fields. X-rays of the skull showed multiple, small, calcified shadows within the sella turcica and also extending for a short distance above. There was very little erosion locally. The changes were thought to be typical of craniopharyngioma (Fig. 3).

Operation. On June 28, 1938 a right frontal osteoplastic flap was turned down. The dura did not appear to be under increased pressure. The frontal lobe was elevated and when the chiasmal region was exposed the yellowish-white dome of a cystic tumor was seen projecting between the optic nerves. The typical, oily fluid of a craniopharyngioma escaped from the cyst when it was opened. The tumor

which was the size of a hickory nut, was removed in one piece. The optic nerves were then well visualized and did not appear to be abnormal. It was believed by the operator that a complete removal had been accomplished. Closure was effected in the usual manner.

Course. The patient made an uneventful recovery and was discharged from the hospital on July 8, 1938.

She was seen last on Nov. 17, 1947. She was of normal height and weight. There was no polyphagia, polydipsia or polyuria. The menarche had begun 19 months before this out-patient visit and the periods had occurred regularly every 30 days. Secondary sexual characteristics were normally developed. Occasional headaches had occurred which she attributed to reading and studying too much. There were no complaints of blurred vision, visual field loss or diplopia. At this time she was doing very well in school in the 7th grade, carrying an all "A" record.

Check-up x-rays of the skull obtained at this time showed the changes incident to osteoplastic craniotomy of the right frontal bone but the sella appeared normal and there was nothing to suggest recurrence.

Microscopical Diagnosis. Adamantinoma.

This case presents several important aspects. The tumor was small, cystic and easily enucleable in one piece. Certainly one should not be content with less than a total removal in this situation. The postoperative course of this patient exemplifies the result that may be obtained from this approach. At the present time, 10 years after operation, the patient is well, of normal height and weight and is menstruating normally.

Case 3. E.L., a $2\frac{1}{2}$-year-old child, was admitted on Sept. 8, 1947 with a chief complaint of blindness in both eyes. In January, 1947, 9 months before admission, his parents noticed that when he was placed near a lamp he would cover his eyes. This also occurred when he was outside in the snow or sun. Photophobia continued and he appeared to be gradually losing vision in the left eye. About the middle of June, 1947, he began stumbling over objects when walking and bumped into walls. He would feel about for toys and had all the appearances of being unable to see. Around July 30 the child became irritable and would cry frequently. He began to regurgitate after taking food. This situation continued and for 2 weeks previous to admission he was more irritable, and had increasing fatigue and frequent regurgitation. For the last 1–2 months he had had polydipsia and polyuria.

Examination. He was a well-developed, well-nourished, slightly obese child who was obviously unable to see. His height was 31 inches and his weight 30 lbs. Blood pressure was 92/50.

The pupils were dilated and reacted poorly to light. A divergent, alternating strabismus was present. There was no nystagmus. There was pallor of both disks. Reflexes were symmetrically diminished. There were no paralyses. There was a questionable Babinski sign on the left. Laboratory findings were normal.

X-ray of the skull showed a rounded shadow of calcium lying immediately above the sella turcica, and in the midline (Fig. 4). There was no evidence of erosion of the anterior or posterior clinoids. There was no significant enlargement of the sella turcica. The sutures were not widened and there was no increase of digital markings. X-ray of the long bones showed no convincing evidence of abnormality.

Operation. On Sept. 22, 1947 a right frontal flap was turned down and the dura

exposed. The dura did not show evidence of any great increase in intracranial pressure. The frontal horn was tapped. The dura was then followed along the floor of the anterior fossa until the lesser wing of the sphenoid was identified. The dura was opened at this point and the exploration then carried intradurally. On elevation of the frontal lobe a bluish, calcified, cystic mass was seen which obscured the right optic nerve. The cyst was opened and following its collapse it was possible to see the optic nerve. Both optic nerves had been markedly compressed. The cystic tumor was only slightly adherent to the optic nerves and chiasm. It was teased out from under the left optic nerve and chiasm and then from beneath the right optic nerve. The calcified cyst was then removed in toto.

Course. The immediate postoperative course was somewhat stormy, with tem-

FIG. 4. Case 3. X-ray showing calcification in cyst wall.

perature varying between 101–103°, pulse 100–120, and blood pressure from 120/70 to 90/40. The patient responded slightly but not well. On the 2nd postoperative day he was taking fluids by mouth and talked to his mother. Except for listlessness, his improvement continued. By the 7th postoperative day he was responding well but showed pyramidal signs on the left with spasticity, a Hoffmann on the left and a Babinski sign on the left. He was discharged on Oct. 23, 1947 in good condition except for the fact that he was still totally blind.

The patient was examined in the Out-Patient Department on Dec. 11, 1947. He appeared to be bright and doing very well. He had gained some weight. He continued to show marked diabetes insipidus. It was the opinion of his relatives that he was regaining some vision in the left eye. Optic atrophy was still present as before.

In May 1948 he was seen again. He was asymptomatic except for some diabetes insipidus. Examination by the Opthalmology Department showed there there was definite return of some vision in the left eye.

Microscopical Diagnosis. Adamantinoma.

Case 4. This patient was included in the original series of 3 cases reported by Peet[7] in 1927. R. B., a 6-year-old girl, was admitted on Sept. 8, 1927 with a chief complaint of left-sided headaches for 6 months. Visual disturbance in the left eye had been discovered for which she had been fitted with glasses without relief. The only positive findings on examination were optic atrophy OS with light perception only and temporal hemianopsia on the right. X-rays of the skull showed evidence of increased intracranial pressure, enlargement of the sella turcica, erosion of the body of the sphenoid, and suprasellar calcification. On Sept. 13, 1927 a right frontal osteoplastic flap was turned down. The frontal lobe was elevated and the chiasmal region exposed. There was a smooth tumor between the optic nerves. The left portion of this tumor was definitely cystic. The entire tumor was removed leaving no capsule in the sella. The patient had an excellent postoperative course and was discharged on Sept. 28, 1927.

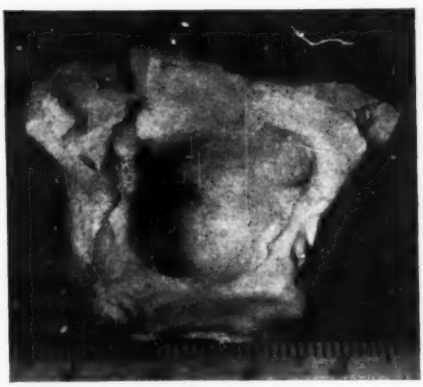

FIG. 5. Case 4. Sella turcica removed post mortem, showing absence of the pituitary gland.

Follow-up History. After discharge from the hospital the patient did well and had no complaints. On Dec. 3, 1931 she was readmitted in convulsions. She had complained of general malaise for 3 days previously and on the morning of admission had begun to have convulsions with cyanosis. On examination tonic and clonic convulsions were noted. CSF cell count was normal. Temperature was 108°. In spite of supportive measures the child suddenly became cyanotic and respirations ceased.

Postmortem Examination. Pathological diagnoses were: acute purulent bronchitis and bronchopneumonia; severe, acute purulent endometritis, salpingitis, pelvic peritonitis and periappendicitis, acute passive congestion and moderate parenchymatous degeneration of all organs; hyperplasia and hypertrophy of adrenal medullae; early adenocarcinoma of thyroid arising in a small proliferation adenoma; acute passive congestion and edema of brain and spinal cord. There was no evidence of a pituitary gland. The sella turcica was removed in a block at necropsy and photographed (Fig. 5). No recurrent tumor was seen.

There was absolutely no pituitary tissue remaining in the sella as can be seen in Fig. 5. This is especially interesting in view of the fact that during the 4-year period prior to her death this patient showed evidence of growth and at the time of death was within 4 cm. of the estimated height for a child of that age. It was also disclosed at postmortem examination that there was normal, functioning ovarian tissue with mature and developing Graafian follicles.

DISCUSSION

A goodly proportion of the craniopharyngiomas are amenable to complete and successful surgical removal. This is particularly true in very young

children before fixation of the tumor to the optic nerves, carotid arteries and hypothalamus has taken place. A certain number of the craniopharyngiomas, on the other hand, are readily seen to be hopeless from a surgical standpoint because of their large size as demonstrated by x-ray.

It is the cases that do not fall readily into either of these categories that pose such a difficult problem in surgical judgment. It is in these cases that surgical exploration should be made, reserving final decision as to procedure until the region of the chiasm has been exposed. If the tumor is found to be cystic, the presenting wall should be excised. One may then be able to free up the tumor from the optic nerve and carotid artery exposed on the side

FIG. 6. Pneumoencephalogram showing large tumor filling the 3rd ventricle (W. R., aged 8).

of the bone flap. At this stage it is sometimes surprising how moderate traction may result in sudden delivery of the blind end of the mass which has been projecting into and blocking the anterior 3rd ventricle. It should also be mentioned here that only slightly more than moderate traction may result in tearing of the unexposed carotid artery, a most painful experience and one that has happened to us on more than one occasion. This is, however, a type of surgery never lightly entered into, so that risks must be taken.

At the time of the exploration hard and fast rules cannot be given, as so much depends on the experience of the individual surgeon. We have undoubtedly erred on the side of being too radical as is attested by our operative mortality. The point we wish to stress, however, is that too few attempts have been made by competent surgeons to remove these tumors totally.

An example may be given here of a recent case not included in this series as the patient entered the hospital after the date line set for this study. The patient was an 8-year-old boy in whom air studies revealed evidence of an

exceedingly large uncalcified suprasellar tumor (Fig. 6). This would ordinarily have been considered hopeless from a surgical standpoint. Operation was performed, however, but only the anterior third of this solid tumor could be removed. Three months later another attempt was made to remove the tumor. The mass had now extruded itself from the anterior 3rd ventricle and was removed in toto. It was believed that the tumor had migrated in much the same way that an abscess migrates toward the surface after one decompresses over it. Though this boy died 6 weeks postoperatively, it was suggested that some of these more inaccessible tumors might be handled in this way.

X-ray therapy in these cases has been little used in this clinic as it was our opinion that it is of no value.

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THE OCCURRENCE OF HEMANGIOBLASTOMAS (TWO CEREBELLAR AND ONE SPINAL) IN THREE MEMBERS OF A FAMILY

W. McK. CRAIG, M.D., AND GILBERT HORRAX, M.D.

Section on Neurosurgery, The Mayo Clinic, Rochester, Minnesota, and Department of Neurosurgery, The Lahey Clinic, Boston, Massachusetts

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THE familial character of hemangiomatous cysts and tumors of the central nervous system has been stressed by numerous writers, this characteristic perhaps being true particularly of patients having associated angiomatosis of the retina as well as cysts of other organs such as the kidney, pancreas, liver, and so forth, the so-called Lindau-von Hippel disease. Lindau[3] himself believed that about 20 per cent of all cases showed a family incidence.

Although hemangiomatous cysts in the cerebellum with or without similar associated lesions elsewhere are by far the most frequent form of this tumor situated in the central nervous system, nevertheless the spinal cord occasionally has been found to be the seat of hemangiomatous or angiomatous growths. Cushing and Bailey[2] felt that spinal lesions of this nature were extremely rare. However, Sargent[5] stated that he had operated upon 6 patients with spinal cord angiomas, and Wagener, Kernohan and one of us (W.McK.C.)[1] reported several cases of typical hemangioblastoma of the spinal cord in which no other lesion was clinically demonstrable. One of these patients had histologically verified cerebellar and spinal cord hemangiomas, and from his history it is evident that his disease was familial, although it was not possible to verify the lesions in other members of his family except for an angioma of the retina in 1 sister. In 1932 Dorothy Russell[4] reported a case of capillary hemangioma of the spinal cord associated with syringomyelia, noting the great rarity of the condition.

The 3 patients to be reported here present several interesting features. In the first place, the hemangiomatous cysts of the cerebellum in the mother (Case 1) and 1 daughter (Case 2) have been verified histologically. The mother likewise had a probable angioma of the retina (and kidney?) and was reported previously by one of us.[1] One daughter (Case 2) had two separate verified hemangiomas, one in each cerebellar hemisphere. The other daughter (Case 3) has angiomatous or hemangiomatous tumors, identified by inspection, involving both the cervical and lumbar portions of her spinal cord.

Case 1. Slowly developing cerebellar symptomatology with retinal hemangioma difficult to recognize. Ventriculography with removal of hemangioma in the left cerebellar hemisphere. J.F., aged 42, was referred to the Mayo Clinic in September 1937 for examination. She had had some loss of vision in the right eye for 11 years which had

been diagnosed as chorioretinitis with scotoma. Recently intermittent headaches and vomiting had developed.

Family history was negative except for questionable migraine headaches in the mother.

Present Illness. The patient noticed difficulty in vision of the right eye which had been present for 11 years with no other symptoms and was considered to be chorioretinitis of tuberculous origin. Because of the headaches, an encephalogram was done, which was negative. She returned in June 1938, with a history that following encephalography there had been some relief of the headaches and very few neurologic symptoms. Four months before re-examination headaches, projectile vomiting, and some unsteadiness of gait had developed.

Examination. The patient was ambulatory, cooperative and fully oriented. There was marked incoordination of the left arm and leg with slight ataxia. Examination of the eyes revealed nystagmus on looking to the right, and papilledema of 4 D. on the right and 5 D. on the left. The left visual field was normal; the right showed a loss of the temporal field and the nasal upper quadrant, with diminution of vision which could have been explained by the retinal lesion. The retina had the appearance of proliferating retinitis with detachments of the lower portion of the retina in the right eye. In view of the development of cerebellar signs, the retinal lesion was reconsidered and the consensus was that it might be an angiomatous lesion of Lindau-von Hippel's disease .

Operation (W.McK.C.). On June 24, 1938, ventriculography was performed with removal of several ounces of fluid. Subsequent roentgenograms revealed dilatation of the lateral and 3rd ventricles with some elevation of the posterior horn of the left lateral ventricle.

A suboccipital craniotomy was done in the upright position under intratracheal anesthesia. When the dura was opened, the inferior tonsil of the left cerebellar lobe was found to be herniated below the level of the 1st cervical vertebra. The convolutions of the left cerebellar lobe were broadened and flattened and the entire lobe was covered with large, engorged, tortuous vessels (Fig. 1, a). A needle was inserted and 0.5 cm. below the surface a cyst was encountered from which 5 cc. of fluid were removed. The fluid was yellow and coagulated on standing. The vessels on the surface of the cerebellum were ligated and coagulated. An incision was made into the cyst and a nodule was found lying on the lateral wall (Fig. 1, b). The cyst wall, including the mural nodule, was removed completely. Convalescence was satisfactory.

Subsequent Course. An examination in March 1945, 7 years after operation, revealed no papilledema and no neurological disturbances. Vision was normal except for the defect due to the retinal lesion of the right eye which had not increased in size.

Pathologic Report. The tumor, which weighed 5 gm. and measured 3×2×2.5 cm., was a hemangio-endothelioma. On microscopic examination it was found to contain more large thin-walled blood spaces than are usually seen in hemangioblastomas. There were regions in which numerous small blood spaces were lined by endothelial cells, some of which were swollen and contained vacuoles that gave the cells a typical foamy appearance. There were, however, more foam cells between the blood spaces than actually lining the spaces. Staining showed the vacuoles of the foam cells to contain fat. The relationship of the xanthic cells to the walls of the blood spaces was best demonstrated with a combination of Perdrau's silver impregnation method for connective tissue and scarlet red staining for fat.

Comment. Like many patients with hemangiomatous cysts of the cerebellum this patient presented a difficult diagnostic problem since only minimal evidences of loss of cerebellar function were present. Even the early encephalographic studies were inconclusive and this has been true of other patients suffering from the same type of tumor. However, there eventually developed intracranial hypertension and cerebellar signs.

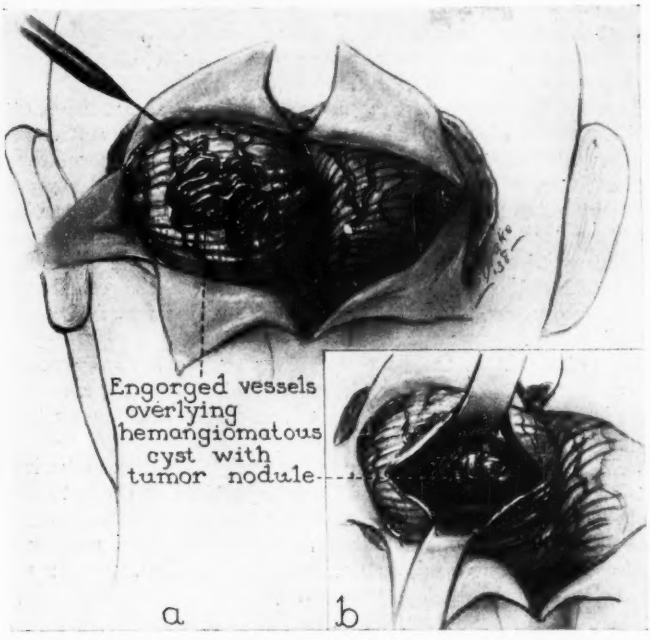

FIG. 1. (From Craig, Wagener and Kernohan[1]) Appearance of surface vessels (*a*) and underlying solid and cystic tumor (*b*) in Case 1.

There can be little doubt that this woman's retinal lesion represents an angioma and the case is an excellent example of an early manifestation of the retinal angioma which in its more chronic phase may be confused with other types of lesions of the retina; in this instance it simulated tuberculous choroidoretinitis with periphlebitis. Only with the development of cerebellar symptoms and intracranial hypertension was the retinal lesion suspected of being von Hippel's disease.

Case 2. Meager cerebellar symptomatology. Dilated ventricular system by ventriculography. Disclosure and complete removal of hemangiomas in both cerebellar hemispheres. E.F., aged 18, was referred to the Lahey Clinic and admitted to the New England Deaconess Hospital on Aug. 16, 1942. She is a daughter of Mrs. F. (Case 1),

and had been complaining of headaches, nausea and vomiting for 2 or 3 months.

Family History. Her mother, as noted, had been operated on by one of us (W.McK.C.) at the Mayo Clinic 4 years previously and an hemangiomatous cyst of the cerebellum removed. Her past history was negative for serious illness or head trauma.

Present Illness. The patient had been well until 2 to 3 months before admission when she began to have headaches which were frontal and occipital in situation. These were dull and of short duration at first, but became severe and almost constant. She had also suffered from occasional morning nausea and vomiting, had become slightly unsteady, and had noticed some numbness of her extremities.

FIG. 2. Case 2. Dilated lateral and 3rd ventricles together with dilatation of upper portion of aqueduct of Sylvius.

Examination. The patient was conscious, cooperative and fully oriented. Her pupils were equal and reacted normally. There was a persistent nystagmus vertically and on looking to either right or left; otherwise the ocular movements were normal. Examination of the fundi showed slight blurring of the nasal margins of the disks with filling in of the optic cups. Visual acuity and fields were normal. No other neurological abnormalities were found other than absent abdominal reflexes. Roentgenograms of the skull were negative.

Lumbar puncture revealed an initial pressure of 400 mm. of water with the patient well relaxed. There were no cells; total protein was 43 mg./100 cc.

In view of the high CSF pressure, ventriculography was carried out on Aug. 19, 1942; 90 cc. of fluid were obtained, and replaced by a similar amount of air. The subsequent x-ray films showed a greatly dilated ventricular system with the upper portion of the aqueduct also visualized and dilated (Fig. 2). Immediately following ventriculography, a suboccipital craniotomy was performed.

Operation (G.H.). The suboccipital region was exposed by the usual curved incision and a wide bony opening secured over both sides.

When the dura had been opened an obvious small tumor was at once apparent on the lower lateral surface of the right cerebellar hemisphere. It consisted of a bright red central portion, some 3 to 4 mm. in two diameters, and surrounding this there was a greyish-yellow area about 1 cm. in size. This whole area was quickly and easily excised completely, using the electrosurgical apparatus, and was preserved intact for pathologic study (Fig. 3).

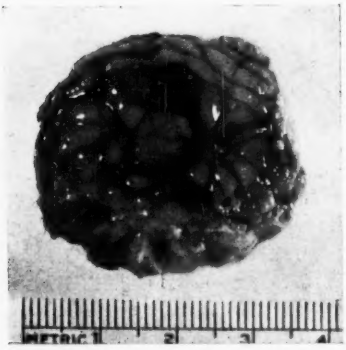

FIG. 3. Case 2. Photograph of small tumor in right cerebellar hemisphere (see Fig. 4). (Natural size)

It was noted that the cerebellar tonsils were herniated far through the foramen magnum and it was thus obvious that the small tumor that had just been removed could not be responsible for this. Upon inspecting the cerebellar hemispheres, the left showed widened convolutions, and toward its lateral portion the convolutions were likewise discolored. A needle introduced into this area secured a few cc. of yellow cystic fluid. The cerebellum was incised down to the cyst and thus exposed a large, solid, red-colored tumor extending from the lateral wall of the cyst outward and upward to the tentorium (Fig. 4). This solid tumor was roughly the size of a hen's egg, and was finally removed completely after an extremely difficult session occasioned by severe hemorrhage.

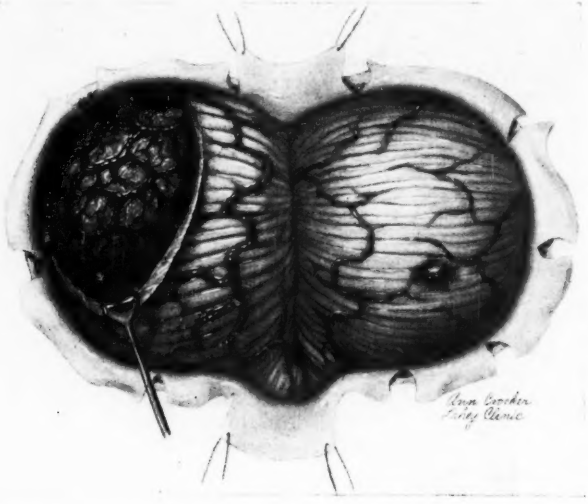

FIG. 4. Case 2. Elaboration of operative sketch of the two cerebellar tumors: small superficial tumor in right hemisphere; large, partly cystic but largely solid tumor of left hemisphere.

The patient made an excellent but somewhat prolonged convalescence and was discharged on Sept. 30, 1942.

Subsequent Course. On Aug. 6, 1947, 5 years postoperatively, the patient was seen for a follow-up examination. She was in perfectly normal health; had been married 1 year after operation and has had 1 child. She had no cerebellar or pressure symptoms of any kind.

Pathologic Report. Microscopic sections from the large tumor presented quite a variable picture. For the most part it was composed of two elements, endothelial-

FIG. 5 (*left*). Case 2. Low power (×90) photomicrograph of the large tumor in the left hemisphere.
FIG. 6 (*right*). Case 2. High power (×710) photomicrograph to show detail of cellular elements.

lined spaces filled with blood, and clusters of mesenchymal cells. The vascular channels exhibited a wide range in size, several being almost a low power field in diameter while the majority were much smaller, of approximately the diameter of liver sinusoids. The larger vessels had a definite fibrous or fibromuscular wall. The smaller vessels were delineated by a single thick membrane against which one or two endothelial cells might be found in each space. In some channels the endothelial cells and membrane appeared to be one unit; elsewhere they appeared to separate (Fig. 5).

The cellular component consisted of small clusters, 2 to 5 usually, of varying-sized clear cells lying between the vascular channels described above. These clusters were surrounded by a thick membrane which, with the small vessel walls, formed a reticulated pattern. The cells themselves had a clear white cytoplasm with hematoxylin and eosin stain or a cytoplasm that was composed of homogeneous tiny

vacuoles. Their nuclei were generally oval or wrinkled, and ranged from approximately 10 to 25μ in greatest dimension and had a sparse amount of chromatin. Mitoses were not seen (Fig. 6).

The histologic picture was that of an hemangioma.

Microscopic sections from the small tumor in the right hemisphere were similar.

Comment. The 2 patients whose records have just been cited, although having almost identical lesions in the left cerebellar hemisphere, nevertheless presented contrasting clinical features. Although the mother (Case 1) had had symptoms 1 year previous to her first neurological survey, an air study failed to show any changes in the ventricular system. On the other hand, her daughter (Case 2) showed great ventricular dilatation after only 2 or 3 months of headaches, and in the absence of cerebellar signs other than nystagmus. The paucity of clinical localizing criteria has been a feature in other patients with similar lesions and emphasizes again the great value of ventriculography as well as its occasional misleading evidence.

The extreme operative difficulties due to hemorrhage from enormous, friable blood vessels, especially in the presence of a large solid portion of an hemangioblastoma, was a common feature of both mother and daughter.

Case 3. Progressive symptoms of spinal cord pressure for 3 years. Disclosure by operations of angiomatous or hemangiomatous lesions at cervical and lumbar enlargements. V.C.F., younger daughter of Case 1 and sister of Case 2, was first admitted to the New England Deaconess Hospital on May 24, 1944, complaining of pain in the back and progressive weakness of the legs for 3 years.

Family History. As noted, her mother and sister had been operated upon previously for hemangiomatous cysts of the cerebellum.

Present Illness. The patient had been well until 3 years previous to admission when she first noted pain in the middle and low back region. This pain gradually increased in severity and radiated around both sides of the trunk at the level of the lower ribs. It had been sufficient to awaken her in the morning. During the past 6 to 8 months she had had pain likewise in the left axillary region running down the left arm, and this pain was increased by coughing.

About 1 year before admission she had first noted some weakness of the legs, particularly awkwardness in running. This weakness progressed so that she had become unsteady and her right leg sometimes collapsed under her. Both legs had felt numb as if she were "walking on cushions."

Neurological Examination. The fundi and other cranial nerves were negative except for complete nerve deafness on the right side which had been present for 4 or 5 years. *Motor System.* There was general weakness of both legs in all muscle groups, but no atrophies. She could stand and walk by herself, but unsteadily and on a wide base. There was slight relative weakness of the left hand as compared with the right and slight atrophy of the intrinsic muscles of the left hand on its dorsal aspect. *Sensory System* (Fig. 7). There was fairly complete loss of pain sensation over the thoracic dermatomes from the 1st to the 8th or 9th level on the left side, and both thighs were analgesic up to the level of the 12th thoracic or 1st lumbar dermatome. Some disturbance of sensation was present over the lower legs and feet, but this was less marked than over the thighs. There was some slight diminution to pin prick over the whole abdomen above the 12th dermatome. In addition, position

and muscle sense were lost in both feet, and both feet were cold. There was marked ataxia in the heel-to-shin test with both legs and a positive Romberg's sign. *Deep reflexes* were active and equal throughout in both arms and both legs. Plantar responses were normal and there was no ankle clonus.

Roentgenograms of the skull and the entire spine showed no abnormalities.

Lumbar puncture was attempted on May 25, 1944. No fluid could be obtained throughout the lumbar area and at the 12th thoracic interspace.

Preoperative diagnosis: Spinal cord tumor (single or multiple). It was felt that the growth might be an intramedullary hemangioma in view of the family history.

Operation I (G.H.). On May 26, 1944 laminectomy was carried out from the

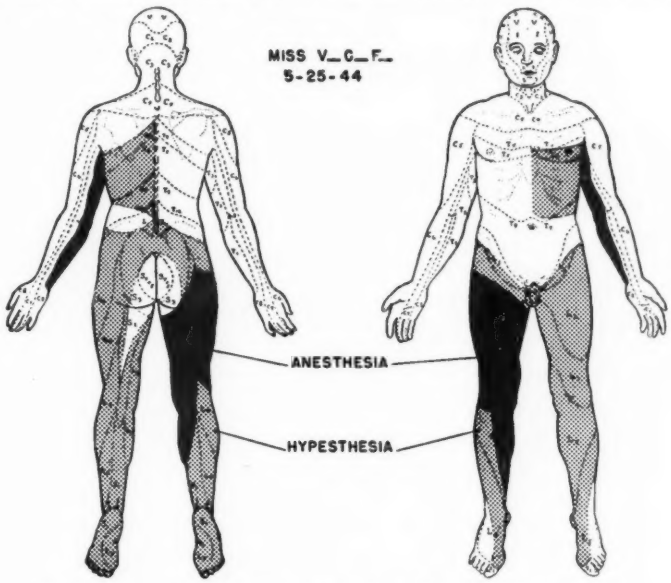

FIG. 7. Case 3. Chart of sensory loss on 1st admission, May 24, 1944.

3rd cervical to the 1st thoracic vertebra. The bone was over-vascular, especially on the left side where the bone was softer than on the right. There was no pulsation of the dura. When the latter was opened throughout the exposed area there was immediate bulging of the cord through the dural incision. The cord seemed to fill out the arachnoid completely so that it gave the appearance of being expanded by a tumor within its substance. The surface of the cord had the appearance of a diffuse angioma or hemangioma, being covered with large, tortuous blood vessels throughout the extent of the area exposed. No attempt was made to deal with these vessels, but the dura was left widely open for decompressive purposes.

The patient made a rather prolonged convalescence and was given a course of 18 roentgen treatments directed to the entire cord.

Nov. 14, 1944—5½ months after operation. The patient came for a check-up examination. She had improved gradually and walked by herself with only slight unsteadiness.

Jan. 7, 1945. She was readmitted to the hospital because of progressive ataxia and loss of position sense in her legs.

Motor Examination. The patient had regained the full strength and use of her hands and arms, was able to sew, knit and carry out all normal movements with her hands. Her legs were weaker and much more ataxic. The heel-to-shin test was done extremely poorly with both legs and there was complete loss of postural sense in both legs. *Sensory Examination.* This showed hypesthesia to all modalities over both lower extremities, but sparing the sacral segments. The area of hypesthesia extended upward over the abdomen to the level of the 12th thoracic segment on the right, and on the left there was lowered sensation up to the 5th cervical segment

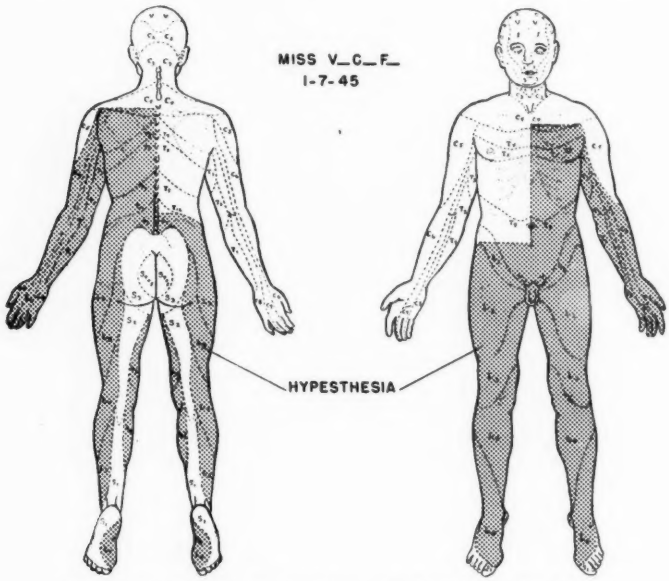

FIG. 8. Case 3. Chart of sensory loss on 2nd admission, Jan. 7, 1945.

(Fig. 8). *Deep Reflexes.* The triceps reflexes were either absent or equivocal on either side, but all other deep reflexes were active and equal. Plantar responses were inactive on both sides.

Lumbar puncture and oxygen myelography were carried out on Jan. 13, 1945. A needle introduced between the 2nd and 3rd lumbar vertebrae encountered slightly xanthochromic fluid. There was a complete subarachnoid block by jugular compression. Subsequent roentgenograms showed that the upper level of the oxygen was at the 1st lumbar level, no gas being present above this. The total protein content of the fluid was 2,515 mg./100 cc.

Operation II (G.H.). On Jan. 16, 1945 a laminectomy from the 9th thoracic to the 1st lumbar vertebra inclusive was performed. The dura when exposed was of a bluish-red color and there was a distinct bulge in the dura at the level of the 10th and 11th thoracic areas. The dura felt firm to palpation. The following is a quotation

from the operative note: "The dura was opened over the area exposed and an extraordinary picture presented itself. Toward the upper end of the dural opening there was a lesion which was bright pink in color and smooth in contour. This pink area covered the cord like a saddle with sharply demarcated upper and lower margins and extended around on either side of the cord as far as one could see. Above and below this area of presumed hemangioma there was a tangle of large and small interlacing blood vessels, and from the lower side one particularly large vessel in the

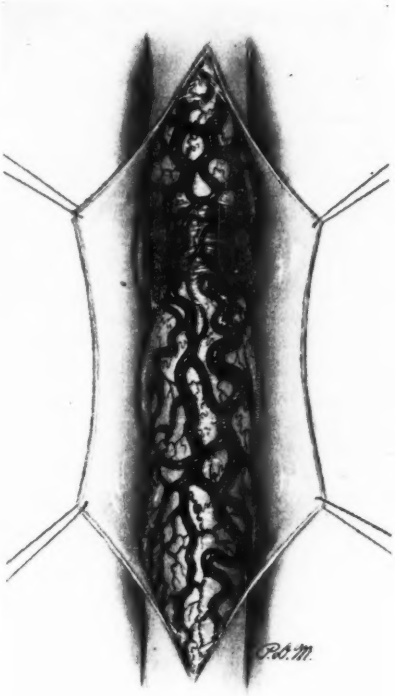

FIG. 9. Elaboration of operative sketch of tumor involving lumbar cord.

form of a corkscrew seemed to come up and enter the under surface of the pink saddle area between the latter and the cord (Fig. 9). By retracting the blood vessel mesh below the pink area one could see what may have been a further extension of tumor anterior to the mesh of vessels, this extension being yellowish-grey in color. A similar appearing area could be seen by retracting the vessels above the pink saddle area. The dura was left open for decompressive purposes."

The patient was discharged on Mar. 12, 1945. She was able to get about in a walking frame. Her legs were very weak and numb and she had marked ataxia.

Check-up Examinations. Aug. 7, 1946—1 year and 8 months following 2nd opera-

tion. In the interim the patient had been given 3 courses of x-ray treatment covering her entire spinal canal by Dr. Lawrence Reynolds of Detroit.

She was now able to walk fairly well, usually with a single cane, but could walk without any support. Her legs had become considerably stronger than they were when she left the hospital, but they felt numb and she had difficulty in knowing where her feet were placed. She had no pain.

Examination showed that her arms and hands were normal in strength and in all movements. Her legs showed fairly good strength and there were no atrophies. *Sensation.* There was a band of hypesthesia to all modalities from the 8th cervical to the 3rd thoracic dermatome inclusive on the left side, and likewise hypesthesia over both legs and both sides of the trunk up to the level of the 6th thoracic on the right and the 8th thoracic on the left. *Deep reflexes* were normal at both elbows, but somewhat overactive at both knees and both ankles. There was normal plantar flexion on both sides.

Aug. 5, 1947—2½ years after 2nd operation. The patient had lost some ground so far as her legs were concerned. A year previously she had reached the highest point in her recovery and had been able to play a few holes of golf.

Examination showed that both hands and both arms were of normal strength without atrophy. The right leg was extremely weak in all muscle groups, while the left leg still retained fair strength throughout. Sensation was about as charted in Fig. 8, except that there was now some slightly lessened sensation over the right side of the upper abdomen and chest up to the level of the 4th thoracic segment. The deep reflexes were present and equal at both biceps, but rather underactive. The knee jerks were likewise present and equal, but less active than normal. The Achilles reflexes were active on both sides, but greater on the left. There was no response to plantar stimulation on either side.

Nov. 15, 1948. The patient was last seen on this date, having been readmitted to the hospital for observation and any possible treatment several weeks previously. At this time it was evident that the weakness of her legs, particularly the right, had become more marked so that she was unable to stand or walk. Her arms and hands showed excellent strength and no sensory loss. The sensory changes over her trunk and legs were about the same, except that the loss now was bilateral up to the 4th thoracic dermatome.

SUMMARY

The 3 patients whose histories have been detailed are reported largely to put on record another instance of the familial incidence of hemangioblastomas of the central nervous system. Certain unusual features were at hand, namely, the presence of tumors in both cerebellar hemispheres in Case 2, and either two separate tumors involving different regions of the spinal cord in Case 3 or else the possibility that the angioma or hemangioma may run throughout the entire length of the cord.

Certain marked differences in the symptomatology and clinical findings in the 2 patients with cerebellar tumors have been mentioned. In one, normal ventricles were disclosed after intracranial pressure symptoms had been present for a year, while in the other there was great ventricular dilatation only 2 to 3 months after such symptoms had appeared.

So far as treatment is concerned, both patients with cerebellar tumors

have apparently been cured by the complete removal of their growths which in both instances, however, was accomplished only after extreme operative difficulties. Operative removal of the spinal lesion or lesions in Case 3 was obviously impossible, but an arrest of the condition by deep roentgen therapy sometimes may be hoped for. In a similar lesion of less wide extent so treated we have seen a return to perfectly normal cord function which is still present 10 years following laminectomy and x-ray therapy.

We are greatly indebted to Dr. James B. Ayer of Boston for his valuable consultations in connection with Case 3.

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CASE REPORTS AND TECHNICAL NOTES

PAIN RESPONSES ON STIMULATION OF THE LUMBAR SYMPATHETIC CHAIN UNDER LOCAL ANESTHESIA

A CASE REPORT*

FRANCIS ECHLIN, M.D.†
New York, N. Y.

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The controversy as to whether or not certain pain impulses from the extremities traverse the sympathetic chain on their way to the spinal cord remains unsettled.

Foerster,[3] Leriche,[6] and others are of the opinion that the sympathetic chain contains sensory fibres from the peripheral parts of the body. Livingston,[7] however, has remarked, "Carefully controlled laboratory experiments indicate that *all* pain impulses from the extremities are carried by fibers which go directly to the spinal cord by way of the posterior roots, without entering the sympathetic chain." J. C. White[13] has come to a similar conclusion and states, "There is no valid evidence that the sympathetic system plays any direct rôle in the central conduction of the painful stimuli."

Abundant evidence collected before[11] and during the past war[1,2,4,5,8,9,10,13] has demonstrated that sympathectomy will usually relieve the pain of major causalgia. White[13] and others believe that a plausible explanation for this relief of pain in causalgia may be the section of efferent rather than afferent sympathetic fibres as postulated by Doupe *et al.*[1]

Walker[12] has obtained pain responses on stimulation of sympathetic fibres through buried electrodes. In a series of patients, after performing a preganglionic thoracic sympathectomy according to Smithwick's technique (section of the chain below the 3rd dorsal ganglion and division of the rami to the 2nd and 3rd dorsal ganglia), he placed insulated stimulating electrodes on the distal (peripheral) segment of the divided chain. Subsequent stimulation caused pain in the homolateral upper extremity, the pain response coming on only after a latent interval.

In the present report an experience is described with direct stimulation (electrical and mechanical) of the lumbar sympathetic chain under local anesthesia.

CASE REPORT

History. Jan. 9, 1948. A.R., a white male, 21 years of age, sustained bullet wounds of the right leg in March 1945. A few weeks thereafter a mid-thigh amputation was performed. Phantom limb pain commenced 1 week later when effects of morphine wore off and had been present since. The character of the pain remained the same since its onset. The phantom leg felt as though it were of normal length. The foot felt everted and the toes acutely flexed. There was no sensation of flexion at the ankle but he had the impression that he could move the phantom toes slightly, and this increased the pain in the stump but not in the toes. The pain was usually confined to the foot under the arch and in the toes. At the arch "it feels like someone is sticking a knife into the foot and in the toes it feels as though they are being crushed in a vice." These sensations were constant but the only time the pain was severe was when the patient was lying quietly and not thinking about anything. There was intermittent twitching

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† 555 Park Avenue, New York 21, N. Y.

of the stump, which flexed involuntarily. No pain was present in the stump except on tightening the muscles, which caused sharp pain in the stump itself. There was no burning element in the pain.

In 1945 the patient had 1 paravertebral block with novocain which gave complete relief for 1 hour. In February 1948, 3 additional paravertebral sympathetic blocks were performed. The first resulted in complete relief of the crushing pain in the toes and some relief of the stabbing pain in the arch for 15 minutes. This was followed by an aggravation of the phantom limb pain. Approximately 1 hour's relief of pain followed each of the 2 subsequent blocks. Intravenous sodium pentothal accompanied by suggestion failed to relieve the patient.

In view of the apparent relief from sympathetic block, sympathectomy was advised, although it was explained to the patient that this procedure frequently did not relieve phantom limb pain and no guarantee of a cure could be provided.

Operation. On Feb. 11, 1948, the right lumbar sympathetic chain was exposed under local anesthesia. The skin, subcutaneous tissues and muscles were infiltrated with 1 per cent novocain. Incision from the tip of the 11th rib to the mid point between the umbilicus and the anterior superior iliac spine was made. The approach was then by a muscle-splitting incision through the external and internal oblique and transversalis muscles. The transversalis fascia was broken through by blunt dissection and the peritoneum with its contents reflected medially until the vena cava was exposed. Very gentle dissection was now used along the medial border of the psoas muscle and what proved to be the 3rd lumbar ganglion was brought into view. Retractors were carefully positioned and electrical and mechanical stimulation of the sympathetic chain was now carried out.

Report of anesthetist who recorded the patient's responses to each stimulation

Patient: A.R.
Complaint: Crushing phantom pain in arch of right foot. Also stabbing pain in arch.
Operation: Right lumbar sympathectomy under local anesthesia.
Procedure: Stimulation of intact sympathetic chain, stimulation of distal (caudad) end of cut chain, and stimulation of proximal (cephalad) end of cut chain.

At the time of stimulation the patient complained of no phantom pain but had a "tingling sensation in arch" and could "feel the big toe."

1. 2 V. 2 sec. on 3rd (?) ganglion—"electric shock down stump." The patient cried out.
2. 2 V. 2 sec. on edge of psoas beside ganglion. No effect.
3. 2 V. 2 sec. on ganglion—pain in hip going down stump to arch of foot. "Bad electric shock."
4. 1 V. 2 sec.—pain similar to #1 and #3, but not so severe.
5. Repeat #4—identical to #4 but electric shock extended into big toe.
6. 1 V. at edge of psoas beside ganglion. No result.
7. 1.5 V. beside ganglion. No effect.
8. 2 V. beside ganglion. No effect.
9. 3 V. beside ganglion. No effect.
10. Tapping on ganglion with blunt instrument—complained of burning pain in stump. "Burning that shot down right into my toes."
11. Repeat #10—"It burned like hell that time. Right into my big toe."
12. Silver clip on chain—"It burned like hell right into my toes." One minute later the patient said, "I still feel some burning in my toes. There was none before."
13. Silver clip 4 mm. proximal to 1st clip—"Pain like electric shock in right hip." "Did it go into your foot at all?" "Just a little bit."
14. Chain cut between the 2 clips—"Pain lasted longer than the rest."
15. 2 V. 2 sec. over 3rd ganglion in distal stump (caudad)—exactly same pain as when intact chain was stimulated, but very mild.
16. Clip on proximal chain fell off. It had apparently been loosely applied. When the clip was replaced, the patient immediately had severe, lasting pain in stump. "Electric shock and burning."

17. 2 V. 2 sec. on cephalad stump—"Ow!!!! Oh Christ Doctor, severe pain—electric shock—burning—most of pain at hip but some in foot."
18. Repeat #17—same results.
19. 2 V. sec. on distal stump—"slight burning in foot."
20. Repeat #19, 2 V. 3 sec.—"Just a little bit of tingling, burning—like an electric shock into foot."
21. 2 V. 2 sec. on proximal stump—"Oh God, don't do that one again. Something like the rest but it hurt ten times as much. Burning electric shock from hip all the way down into toes."

The patient stated that at all times the pain, which always radiated from hip to foot, was more severe in hip. His complaint of pain in each instance followed immediately the application of the stimulus. There may have been a few seconds' latent interval between the complaint of mild burning and application of the stimulus to the distal (caudad) stump of the sympathetic chain.

The patient was now given intravenous sodium pentothal and the 2nd, 3rd and 4th lumbar ganglia with intervening chain were removed under general anesthesia.

Postoperative Course. This was uneventful and following is a note made by me on Feb. 24, 1948: Thirteen days ago a right lumbar sympathectomy including lumbar ganglia 2, 3 and 4 was performed. Preoperatively the phantom foot was in its normal position and moved with the foot in the prosthesis. The 1st postoperative day he says the foot didn't seem to move with the prosthesis when he abducted or adducted the leg. Gradually over a few days the foot moved upward toward the knee but he had a hard time localizing it. In about 1 week the phantom foot felt as though it was at the end of the stump and it has remained there but he has difficulty visualizing it. He is not aware of the toes at all—only of the heel and sole—and it feels as though it is asleep. He has been completely free of all phantom limb pain since the operation. He is free of all pain except some ache in the stump at night when he removes the prosthesis. His stump is larger than before and he can wear only 1 sock covering (instead of the previous 3), if he wants to get it into his prosthesis. He has been walking on the prosthesis for the first time since Christmas and this may explain the mild ache in the stump.

The patient was discharged from the hospital on Mar. 1, 1948, at which time he was free of pain. A letter received from him on June 10, 1948 states: "My phantom limb has receded down to the end of my stump. There have been no sharp pains. I just have the feeling that someone is holding the arch of my foot with slight pressure. Since the operation I did not have the intense pain of the squeezing toes and stabbing in the arch. The only thing I feel is when the weather is damp or when it is raining and then the pain is in the arch of my foot. I am relieved of all pain in my toes."

COMMENT

The experience described in this paper illustrates that it is feasible to stimulate the lumbar sympathetic chain under local anesthesia in selected patients.

In the case reported severe pain referred to the phantom limb was elicited (almost immediately) on electrical or mechanical stimulation of the intact lumbar sympathetic chain. After division of the chain, stimulation central (cephalad) to the point of section caused severe pain whereas stimulation distal (caudad) to this point produced only a mild burning sensation in the phantom foot. Such pain responses, it is believed, may be explained on the basis of afferent discharges of pain impulses in the sympathetic chain. It is difficult, if not impossible, to account for the findings reported on the basis of efferent impulses passing out over sympathetic fibres to stimulate somatic pain fibres in the periphery (as suggested by Doupe *et al.*[1]).

CONCLUSIONS

Further evidence is presented that fibres in the lumbar sympathetic chain play an intimate role in certain pain mechanisms connected with the lower extremities.*

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* Since the above observations were made the lumbar sympathetic chain was stimulated in 2 other patients (under local and sodium pentothal anesthesia), each suffering with severe pain in the foot as a result of inadequate circulation caused by arteriosclerosis. In each case when the sympathetic chain had been exposed the patient was allowed to largely recover from the effects of the sodium pentothal. Stimulation of the intact chain (2 volts, 2 secs., 60 cycle alternating current) caused the patient to cry out with pain. After section of the chain above the 3rd lumbar ganglion, stimulation of the distal segment (caudad to section) with 5 volts for 2 secs. or by crushing, caused no response. Stimulation of the proximal segment (cephalad to section) with 2 volts for 2 secs. or by crushing the chain with a hemostat, caused the patient to cry out with pain. The patient was too drowsy to tell us if he could localize the sensation of pain.

A CASE OF EPIDERMOID (CHOLESTEATOMA) OF THE BRAIN AND CAUDA EQUINA

NAOMI RASKIN, M.D.
Boston State Hospital, Boston, Massachusetts

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In his *Anatomie Pathologique du Corps Humain* in 1829 Cruveilhier[6] described a tumor of the brain to which because of its pearly, glistening appearance he gave the name of "tumeur perlée". In 1838 J. Müller[11] described two more tumors of the same type which he called cholesteatoma, because of the presence of cholesterin crystals. Virchow[14] preferred the name given by Cruveilhier and called them "Perlgeschwülste". Objections have been voiced against both these terms by many authors. Horrax[8] called them meningeal cholesteatomas and specified them by a division into the hair-containing and non-hair-containing cholesteatomas. Cushing[7] referred to them as epidermal and dermal cholesteatomas, Critchley[5] gave his reasons for calling them epidermoids, Bostroem[4] divided them into pial epidermoids and pial dermoids, etc. It has been stated by many authors that even with these qualifying terms the name cholesteatoma is unfortunate and that these tumors should be called epidermoids of the brain, if they contain ectodermal structures only, or dermoids, if they are formed of both epidermal and mesodermal derivatives; and that these terms should be used for either intradiploic or intracranial tumors in contradistinction to the so-called cholesteatomas associated with otitis media desquamativa.

These epidermoids are slow-growing tumors, are considered benign and noninvasive, are seldom suspected clinically, and are usually found accidentally either at operation or at autopsy. They are rare tumors, comprising less than 1 per cent of all the intracranial growths. To date, less than 200 cases have been reported in the literature. Therefore, a case of an epidermoid tumor causing clinical symptoms for 17 years and invading the brain tissue seems worthy of presentation.

CASE REPORT

B.S.H. No. 42927. F. C., a 51-year-old white male, was admitted to the Boston State Hospital in May, 1944, with the diagnosis of Psychosis with Convulsive Disorders (Epilepsy), Idiopathic. He began to suffer from epileptic seizures at the age of 34. His spells were of a grand mal type. As his attacks became more frequent, he became confused, talkative, denuditive, suspicious and belligerent.

In 1943 he had been treated for fracture of the frontal bone, lacerated wound of the forehead, contusion of the brain and meningitis, following a fall from an 18-foot wall. He had had ventriculostomy; ventricular and spinal fluid showed normal protein.

He was a well-nourished man. Pupils were regular and equal, and reacted to light and in accommodation. Hearing was poor in the right ear; the right drum was perforated. Reflexes were less active on the right than on the left; the left knee-jerk was hyperactive. Vibratory sensation was present, and there were no tremors. Eyegrounds were negative. Hinton reaction, spinal fluid, blood and urine were negative.

He complained of headaches, had a mild aphasia, continued to have severe epileptic seizures, and died in July, 1946.

Autopsy. The head showed no abnormal configuration. There was a linear healed fracture of the frontal bone, and in the occipital bones there were 2 symmetrical burr-holes, 1.5 cm. in diameter, in the region of which the dura mater was adherent. The pia-arachnoid was thin

and transparent. The brain weighed 1320 gm. The convolutional pattern of both hemispheres showed no abnormalities.

In the right lateral recess, covered with thin transparent pia-arachnoid, there was a tumor of a peculiar shining appearance extending forward along the ventrolateral surface of the pons, covering and enveloping the 9th, 8th, 7th and 5th cranial nerves, to the bifurcation of the basilar artery. It extended backwards along the lateral surface of the right cerebral peduncle to the upper surface of the right upper cerebellar lobe and burrowed itself between the middle cerebellar peduncle and the cerebellum. The tumor was firm but not hard, had a peculiar resiliency, and was very brittle. It had a yellowish, brilliant, and smoothly nodular surface. It appeared to be formed of granules of waxy matter covered with a delicate membranous capsule, which could be easily split into very thin flaky layers. The 5th nerve was stretched over the tumor and at first glance appeared to emerge from its surface, rather than from the surface of the pons (Fig. 1). Externally to the right optic tract and bridging over it

FIG. 1. Epidermoid tumor in the right cerebellopontine angle. It is covered with the pia mater and has a shiny smoothly nodular surface.

was a thickened basal pia which appeared grayish-yellow, granulated, and opaque. The interpeduncular leptomeninges were thickened at the right. The cerebral vessels were supple and there was no evidence of arteriosclerosis. There were several *plaques jaunes* on the orbital surfaces of the frontal lobes, near the frontal poles, and in the middle of the left third temporal convolution. Frontovertical sections through the temporal lobes revealed that the tumor had nestled deep into the hippocampus, for a distance of 0.6–0.9 cm., compressed and destroyed a large part of Ammon's horn, and covered and compressed the lateral surface of the right cerebral peduncle. It had reached the wall of the temporal horn of the right lateral ventricle, but stopped close to it without breaking through the ependyma, which appeared intact. The right choroid plexus looked paler than the left. The ventricular system was free. Sections through the pons and cerebellum showed that the tumor gradually tapered backwards as it wedged between the middle cerebellar peduncle and the right cerebellar hemisphere (Fig. 2).

Sections of the tumor showed it to be composed of a granular, gritty, flaky material which crumbled easily. It was of a dull, pale yellow color in contrast to the brilliant color of the capsule. There were no hairs. The color of the capsule and the granulated structure led to the diagnosis of cholesteatoma or pearl-tumor at the time of the autopsy.

Microscopical Study. The diagnosis was cerebral epidermoid, the term we prefer to that of cholesteatoma. Fig. 3 shows the extent of the invasion of the cerebral tissue, the transition from the pia-arachnoid into the tumor tissue, and the thick border of glia between the tumor and the invaded brain tissue. The thickness and abundance of glial tissue are evidence of a slow invasion of the brain by the tumor. The external layer (the stratum durum, according

FIG. 2. Section through the peduncles shows the cut surface of the cholesteatoma, which is wedged between the right peduncle and the cerebellum. It is composed of separate nodules and can be traced upwards to the inner surface of the pia, where it originated.

to Bailey) was very thick, composed of many layers of wavy collagen fibers, without any evidence of cellular formation. This layer was detached from the underlying granular layer composed of closely packed epithelial keratin-containing cells. This layer was also broken off the deeper layers which formed a mass of cells mostly oval in shape, in which nuclei could not be detected. The desquamated epithelial cells formed the bulk of the tumor. The tumor was completely avascular.

Spinal Cord. Sections of the cord revealed no abnormalities, but attached to the right laterodorsal surface of the cauda equina was a thin shining white tumor, measuring 1.2×0.6 cm., consisting of small granules and covered with a shining thin capsule. The microscopical structure of this tumor was identical with the epidermoid of the brain.

DISCUSSION

Remak in 1854[13] was the first to suggest that these tumors may owe their origin to misplaced epithelial tissue. Love and Kernohan,[9] Bostroem,[4] Critchley and Ferguson,[5] Cushing,[7] and Rand and Reeves,[12] classify them as epidermoids or der-

moids. Epidermoid may be intradiploic, or free in the leptomeninges, or very rarely in the frontal lobes. If located in the diploe, it may be discovered by roentgenography,[1] but situated deep in the brain, it can reveal itself only by the symptoms of an increased intracranial pressure. According to Bailey,[2,3] and Love and Kernohan,[9] the epidermoid tumor is composed of four concentric layers, the acellular stratum durum, which is the cause of the pearly sheen that was responsible for its original

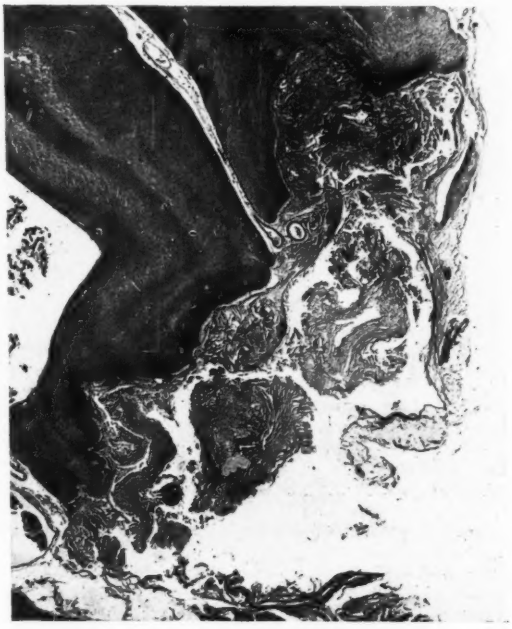

FIG. 3. Photomicrograph (phosphotungstic acid hematoxylin stain, ×13). On the right is seen the capsule of the cholesteatoma which has compressed, destroyed, and invaded Ammon's horn and the hippocampus. The dark pigmented border of the invaded tissues is due to a thick wall of fibrous glia. On the extreme right the intimate relation of the pia and cholesteatoma is seen.

name of pearly tumor, the cellular stratum granulosum containing keratohyaline granules in its cells, the stratum fibrosum, and the stratum cellulosum. Cholesterin is not a constant finding, and when present is due to the breaking down of keratin. By this definition our case belongs to the group of epidermoids that are most frequently found at the base of the brain, their common site being the cerebellopontile angle. The presence of a neoplasm, benign though it may be, is irritating to the brain; and although localizing signs may be missing until the pressure on the neighboring structures gives rise to symptoms, the patient's personality suffers. Most authors comment on the change of personality, mental dullness, frank psychosis and convulsions. In 1936 Mahoney[10] collected from the literature and tabulated 142 cases, 112 of which were intracranial. Of this last group 57 per cent of the patients were psychotic.

Our patient carried the diagnosis of "Idiopathic Epilepsy"; he had had convul-

sions from the age of 34 until his death at 53. He was always considered unstable, nervous, difficult to get along with, and he was frankly psychotic and required hospitalization for the last 12 years of his life.

The epidermoids are said to compress but not invade the brain, but Benda, Cushing and others have described invasion and destruction of brain tissue by these tumors. Our case belongs to this group. The tumor had invaded the hippocampus and had destroyed a large part of Ammon's horn (Fig. 3).

The intradiploic tumors have been removed with success, but the intracranial tumors located at the base, intimately connected with the cranial nerves, may be very difficult to attack. In our case the tumor had enveloped the 9th, 8th, 7th and 5th nerves and extended forward to the right optic tract.

CONCLUSION

1. A case of epidermoid tumor of the brain and cauda equina in an epileptic, psychotic individual is described.

2. The presence of psychosis in patients with these rare tumors is high.

3. As rare as these tumors are, the possibility of their presence in an epileptic, with late appearance of epileptic seizures, should be considered in the differential diagnosis.

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TRAUMATIC BILATERAL ABDUCENT AND FACIAL PARALYSIS WITH GOOD RESTORATION OF FUNCTION

A CASE REPORT

W. Edward Storey, M.D., and William G. Love, M.D.
Surgical Service, Columbus City Hospital, Columbus, Georgia

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Unilateral paralysis of the 6th or 7th cranial nerve is encountered from time to time in disease or injury. Bilateral weakness or paralysis of the 7th nerve is seen in myasthenia gravis, in association with polyneuritis following diphtheria, in polyneuritis of the so-called infectious type as well as in alcoholic B-avitaminosis.[2] However, bilateral facial paralysis of traumatic origin is much less frequent as indicated by various reports. Among 71 cases of closed head injury, Turner[3] reports bilateral facial involvement in 1; none had associated abducent injury. Friedman and Merritt[1] reviewed the records of 430 patients with acute injuries of the head and found involvement of 22 cranial nerves in 19 cases (4 per cent of total). The facial nerve was injured in 7 cases, in 1 of them bilaterally and in association with damage to the left 6th nerve. Therefore when bilateral abducent and bilateral facial paralysis were found following trauma, the case was believed to warrant reporting.

CASE REPORT

A white female of 26 years was rendered unconscious by a blow to the left mandible during an automobile accident on 26 July 1947. There was a slight amount of bleeding from both external auditory canals, chiefly the right. Upon return of consciousness, 12 hours after injury, there was diffuse, severe headache and vomiting. These diminished progressively and ceased by the end of the 2nd day. Upon regaining consciousness, she complained of diplopia and it was noted that her facial expression did not change.

Roentgenograms of the mandible showed a fracture in the region of the left mental foramen with slight upward displacement of the proximal fragment. Views of the skull showed nothing clearly indicative of fracture.

Examination. She was a pleasant, intelligent and cooperative young woman of good development and satisfactory nutritional status. Her recent health included no illness or previous injury. A few small blood clots were found in the right external auditory canal. The neck was not stiff, and no ecchymosis or abrasions were found anywhere. Blood pressure was 116/58; pulse was 64, regular and of good volume; temperature was 99.2 F. Blood count, urinalysis and Kahn test were normal.

A careful neurologic survey (cranial nerves, spinal nerves, cerebellar functions, and autonomic system), made approximately 40 hours after injury, yielded the following pertinent data:

(1) There was bilateral internal strabismus and attempts to abduct either eye past the mid-point failed although the opposite internal rectus moved normally. Therefore, no impairment of conjugate deviation could be said to exist.

(2) The face was expressionless and there was total inability to close the eyes, smile or purse the lips. No thalamic or synkinetic effects could be elicited.

(3) Tests of taste, repeated upon separate days, indicated loss on the right side (anterior $\frac{2}{3}$) but good preservation on the left.

(4) There was no impairment of hearing as tested by tuning fork, pocket watch, or spoken voice. Weber's test was not lateralized and conduction by bone was equally good on the two sides. There was no spontaneous nystagmus on movement of the eyes and no vertigo. Rotatory tests yielded normal results in past-pointing and gait.

Subsequent Course. A week after injury, the patient was free of headache and other generalized symptoms of the head injury and was allowed to return to her home in another city.

At 7 weeks after injury (12 Sept. 1947), re-examination disclosed that the internal strabismus, previously bilateral, persisted only on the left and to a lesser degree than formerly. There was evident improvement in voluntary abduction of each eye but, as yet, maximal abduction was not possible. Hearing continued unimpaired for ordinary conversational tones and vestibular function was normal. Both conjunctivae were slightly hyperemic, the right more so. She volunteered that while she felt all the emotion associated with crying, no tears came from her eyes.

Facial function showed little if any improvement. She was still unable to close her eyes and there was no blinking. Taste could not be retested because of the wired mandible. Hearing seemed normal and all other neurologic functions were satisfactory.

At 9 weeks after injury she began to note impaired hearing and at 10 weeks (3 Oct. 1947) re-examination showed the same findings in the abducent and facial tests as at 7 weeks. The conjunctival hyperemia was less since use of a simple antiseptic-emollient locally. The mandible had been unwired and taste was beginning to return on the right.

At this time the consulting otologist, Dr. John B. Thompson, reported: "Examination of ears reveals both tympanic membranes retracted to a marked degree. Bone conduction is greater than air conduction bilaterally. Schwabach test is increased and Weber's test is lateralized to the right. Audiogram shows impaired hearing with loss of 60 decibels on right and 45 decibels on left in conversational range. Caloric tests on each side, with 10 cc. ice water, show normal labyrinthine reactions. Impression: Air-conductive deafness bilaterally, greater on right, due to hemorrhage in tympanic cavities and, at present, adhesions causing fixation of ossicles and retraction of tympanic membranes."

At 5 months (17 Dec. 1947), she reported that return of facial function commenced about 3 months following injury and shortly after the previous visit. This was first noted in the left cheek. Examination showed ability to close both eyes, wrinkle the forehead, to smile and to purse the lips, although these functions were somewhat weaker on the right. Particles of food frequently accumulated between the right gum and cheek and it was necessary to dislodge them digitally. No diplopia remained. Abducent function and taste were fully recovered. After weeks of eustachian insufflation hearing was somewhat better for conversational tones but no audiogram was made. Otherwise she was well and her morale was greatly improved.

At 13 months (1 Sept. 1948), her chief complaint was a lack of suppleness in the musculature of the right side of the face. It was still necessary, occasionally, to dislodge small particles of food from the right cheek. Examination indicated improved strength in both sides of the face with the right side still weaker. As she spoke, involuntary associated movements were noted as slight winking first on one side and then the other. Abducent function and taste were normal. Hearing was adequate for ordinary conversation but a watch, while audible at 11 inches on the left, was barely heard at 1 inch on the right. She remarked that since April 1948 tears had appeared under appropriate circumstances, but, thus far, they were freer on the stronger left side. This indicated that lacrimal function began to recover at about 9 months after injury and was essentially normal at a year.

Throughout convalescence, no treatment was given beyond hopeful encouragement and attention to nutrition and other general hygienic measures.

COMMENT

In our opinion, the most likely cause of the disability in this case was bilateral basal fracture of the skull. The aural hemorrhage on both sides makes this highly probable, notwithstanding the lack of satisfactory roentgenologic evidence. The bilateral abducent paralysis is accounted for by the fact that this nerve courses over the tip of the petrous portion of the temporal bone on its way to the optic fissure and

was undoubtedly implicated in the fractured area on each side. The initial absence of taste on the right side of the tongue points to injury of the facial nerve at or distal to the geniculate ganglion but proximal to the chorda tympani on that side. Lack of similar impairment on the left side, as well as escape of cochlear and vestibular fibers of each acoustic nerve were assumed to have been fortuitous. Later reduction of hearing was not neural in type but air-conductive and was presumed to be due to adhesions and retraction subsequent to the initial hemorrhage into the middle ears.

Turner[3] states that traumatic facial paralysis is probably always associated with fracture in the petrous portion of the temporal bone, though not always demonstrable by roentgenogram. He believes that facial paralysis developing immediately is most probably due to laceration of the 7th nerve in the fallopian aqueduct. When it appears later, he thinks it is due to the pressure of blood upon the nerve in the canal. In his single bilateral case, paralysis appeared later. In this case, it was evident upon return of consciousness at 12 hours, and was assumed to have been of the immediate type. Aural bleeding and bone-conductive deafness were common among Turner's cases.

At first it was thought that, whether or not basal fracture had occurred, the cause of the neurologic signs might be a transmitted injury to the pons, in the midline, at the level of the colliculus facialis in the floor of the 4th ventricle. This was a convenient site for a single lesion to involve both abducent nuclei and both facial nerves. It spared the assumption of a basal fracture extending bilaterally to involve both facial and both abducent trunks with only right-sided implication of taste and no cochlear or vestibular damage. It would not, however, explain loss of taste on the right side of the tongue. Nevertheless, it was an attractive hypothesis except for the absence of paralysis of conjugate deviation of the eyes. A lesion at or near the 6th nucleus would, in all events, be expected to show this sign. Moreover, it was thought to be likely that a pontine lesion of sufficient degree to involve both abducent nuclei and both facial nerves would present additional features and would prove fatal.

SUMMARY

(1) A blow to the left mandible resulted in bilateral abducent and facial paralysis with involvement of the chordal taste fibers on the right side. There was no demonstrable damage to the cochlear or vestibular portions of either auditory nerve.

(2) There was a slow but progressive improvement in the neurologic defects.

At 9 weeks after injury, bilateral air-conductive deafness was first noted. This was attributed to adhesions and retraction of the ossicles, sequel to hemorrhage into the middle ears.

At 10 weeks, there was substantial improvement in strength of both external recti but diplopia was still annoying. Taste was returning to the affected half of the tongue. Paralysis of the muscles of the upper and lower halves of the face persisted.

At 9 months, lacrimation, which had been markedly reduced since injury, began to return and at 12 months it was essentially normal.

At 10 months air-conductive hearing was still impaired but the abducent defects were entirely cleared. Facial strength was greatly improved though appreciably weaker on the right.

(3) Improvement occurred without efforts at treatment other than hopeful encouragement and attention to general hygiene.

(4) The location of the lesion which would produce the symptoms presented by

this patient is discussed and it is concluded that the nerves were damaged by fracture of the base of the skull.

Dr. William A. Smith, Atlanta, Georgia, and Dr. H. Houston Merritt, New York, N. Y., gave helpful criticism.

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AN AXIAL-ROTATING TABLE

William A. Nosik, M.D.*
Cleveland, Ohio

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There is presented here an axially rotating table which has been designed to facilitate certain neurosurgical and neuroroentgenological procedures.

The prime advantage in the use of this table is the ability of the surgeon to present either side of the head for operation by rotating the patient on his long axis without disturbing the sterile draping about the surgical field. Since the patient and

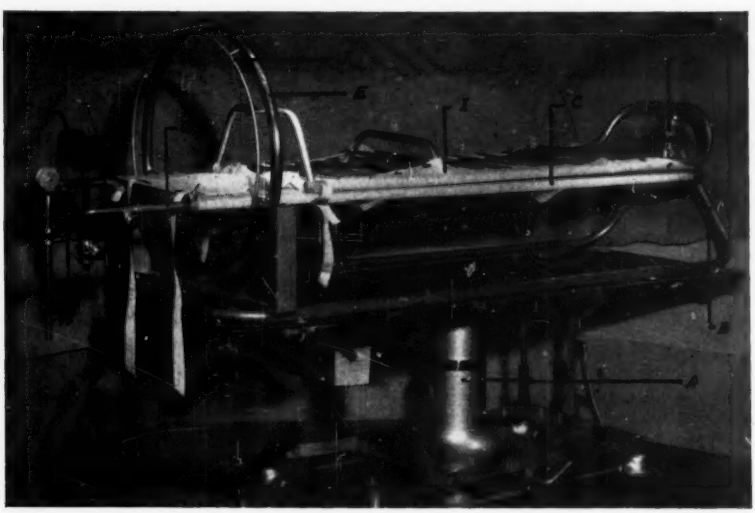

Fig. 1. View of axial-rotating table mounted on movable hydraulic lift.
(For explanation of lettering, see text.)

* 10515 Carnegie Avenue, Cleveland 6, Ohio.

drapes rotate as a unit, the possibility of contamination is lessened. This is particularly true in ventriculography, where the head is often manipulated to gain a more effective emptying of the ventricular system. This table is mounted either on a standard operating table or, as we prefer, on a hydraulic lift-table for convenience

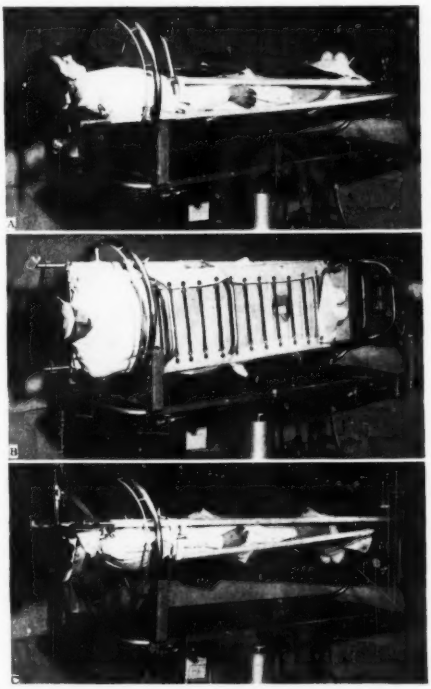

FIG. 2. Patient (A) in position on table, (B) being rotated at 90° and (C) rotated at 180°.

in transporting the patient to the x-ray room. The hydraulic pump is necessary to adjust the unit to the operator's height. Figs. 1 and 2 illustrate how the unit is mounted on the lift (Fig. 1 A), its component parts, and its mode of operation.

The unit (Fig. 1) consists essentially of a supporting frame (*B*) and a movable frame (*C*). This movable frame is so placed that the foot end pivots on the frame at *D* and is welded to a large ring (*E*) which moves on rollers affixed to this support. A locking device is incorporated so that the patient may be securely held in any one of the positions occurring in 30° intervals around the entire ring. The patient's head is secured in a head rest (*F*) which is adjustable in the vertical direction. It is also adjustable in the horizontal direction or capable of being entirely removed by virtue of the telescoping U-shaped bar (*G*).

Another removable telescoping segment (*H*) has an adjustable pulley for those situations where constant traction is desired on the head throughout a procedure in the cervical region. The patient is locked in position on the lower frame by a removable upper frame (*I*) which is bolted to it at the foot end and secured further by appropriate straps.

The patient is draped after suitable preparation of the head by slipping a sterile cone-shaped drape over the head to be held in position by a draw-string or elastic in the groove

behind ring *J*. The size of the exposed operative field may likewise be regulated by drawstring. In the operative preparation, the instruments are placed on a table behind the ring *E* with the drape extending over the sterile cone. In this manner, it is possible to rotate the patient on his long axis without disturbing any of the table drapes. As evidenced in Fig. 2, the patient can be maintained in any desired position of rotation. An effort was made to keep the patient's center of gravity as close to the axis of rotation as possible, thereby facilitating the ease of rotation.

COMMENT

As previously stated, this unit is designed to facilitate certain neurosurgical and neuroroentgenological procedures, particularly where air studies are concerned. In the performance of ventriculography, the field is more adequately presented to the surgeon. With the introduction of the ventricular cannulae, the patient can be rotated through an entire 360° and the ventricles more effectively drained without any additional manipulation of the head. Upon completion of the drainage and the air injection, a dressing is applied and the patient removed to the x-ray room. There the head piece (*F* and *G*) is removed and the head placed on a horizontal Buckey. With the head resting on the Buckey, the patient may again be rotated through any desired arc to obtain any desired angle view of the ventricular system. Since the lateral rotation of the head occasioned by the shoulder is eliminated, it is believed that a truer lateral representation of the ventricular system can be obtained with considerably less effort. Should further surgical procedure be necessary following ventriculography, the patient must be transferred to the standard operating table or chair. This is an acknowledged disadvantage. It is believed, however, that this disadvantage is off-set by the facility with which the air can be injected and the subsequent ventriculograms made.

The application of this unit to trephination as for subdural hematoma, etc., is apparent. With this unit, drainage can be effected by gravity if desired. The operation is shortened by obviating the necessity of re-draping where the bilateral procedure is done.

In certain situations that have been encountered, particularly the traumatic in the cervical region, where it was desirable to maintain a constant head traction while a cervical laminectomy for decompression purposes was done, it is believed that this unit would have facilitated matters. In this situation, the Crutchfield tong is applied and unit *H* attached to the head piece. The weight is then attached to the tongs over the pulley thus maintaining constant traction. The forehead is supported by the head piece, enabling the operator to expose the cervical region. Obviously a modified drape is necessary in a situation of this type.

This unit could likewise be used in making special roentgenographic views of the skull and cervical regions in a patient with a diagnostic problem.

CONCLUSION

An axial rotating table for neurosurgical and neuroroentgenological usage, its component parts and its mode of operation is presented here.*

I wish to acknowledge with appreciation the efforts of Mr. Buehning of the Electromechanical Shop, Cleveland Clinic Foundation, as well as the suggestions of other members of the Neurosurgical Staff of the Cleveland Clinic.

* This unit may be purchased from the Orthopedic Frame Company, 409 East Michigan, Kalamazoo, Michigan.

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